Screening for spina bifida cystica

A cost-benefit analysis

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Hagard, S., Carter, F., and Milne, R. (1976). *British Journal of Preventive and Social Medicine*, **30**, 40-53. **Screening for spina bifida cystica: a cost-benefit analysis.** The costs and economic benefits are examined of introducing a programme for the mass-screening of pregnancies for the detection and abortion of fetuses with spina bifida cystica. A benefit-cost index is derived, and the possible effects on it of making different input assumptions are discussed. It is considered that, on economic grounds, screening may be worthwhile only in populations in which the incidence of spina bifida is high.

In the British Isles spina bifida cystica is among the most common seriously handicapping conditions present at birth. It is responsible for a high proportion of stillbirths and neonatal deaths; even more seriously, it gives rise to substantial family distress, particularly in connexion with the morbidity afflicting many of the survivors (Hare *et al.*, 1966; Freeston, 1971; Walker, Thomas, and Russell, 1971; Hunt, 1973; Richards and McIntosh, 1973; Woodburn, 1974). The considerable economic burden of caring for spina bifida sufferers has not passed unnoticed (Lightowler, 1971; Lorber, 1972).

Means of reducing the burdens upon individual families and upon society as a whole have been proposed. There have been calls for replacement of nearly universal operation by strict criteria of selection of neonates for surgery, in order to produce fewer severely handicapped survivors (Lorber, 1971, 1972); there are indications that selection policies are now widely accepted (Hide, Parry Williams, and Ellis, 1972; Stark and Drummond, 1973; Lorber, 1973). Their practice can be expected to result in a significant reduction in the overall social and economic burdens of caring for those born with spina bifida.

Recently, antenatal diagnosis, enabling selective termination of affected pregnancies, has become

feasible. Brock and Sutcliffe (1972) reported an association between high alpha-feto protein (AFP) levels in the amniotic fluid and anencephaly. A similar association has since been demonstrated for open myeloceles which are responsible for about 90% of serious cases of spina bifida (Allan *et al.*, 1973; Nevin, Nesbitt, and Thompson, 1973). The diagnostic accuracy of the test throughout the second trimester of pregnancy is very high. However, the population incidence of spina bifida cystica does not reach 0.5% in any community, while antenatal diagnosis, since it necessitates amniocentesis, is costly and carries some risk to fetal and maternal health. The practical usefulness of the test is, therefore, restricted. It is currently offered to women who are considered to be at higher risk of bearing a child with a neural tube malformation, chiefly, therefore, to those who have previously borne one or more affected children and thus run about 10 or more times the risk of other members of the population (Richards, McIntosh, and Sweeney, 1972). However, even near universal acceptance of antenatal diagnosis by this higher-risk group would still achieve only a 10% reduction in the prevalence of spina bifida at birth (Lancet, 1974). Its achievement would, in addition, require a substantial commitment of resources.

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In 1973 and 1974 a number of workers reported an association between raised AFP levels in the maternal serum and the presence of a fetus with a neural tube malformation (Leek *et al.*, 1973; Brock, Bolton, and Scrimgeour, 1974; Wald, Brock, and Bonner, 1974).

Since these developments have given rise to hopes of offering routine screening in pregnancy for the detection of spina bifida, it is appropriate to consider the conditions which need to be met (Cochrane and Holland, 1971; Chamberlain, 1973) before current hopes can be realized.

The screening test should be simple, acceptable to the subjects, accurate, and repeatable. The extent to which it meets the ideal of establishing either the presence or absence of disease in every individual screened should be known. In screening for spina bifida, serum AFP estimation would be performed on blood taken at an ordinary antenatal visit—a simple, acceptable, routine procedure. Current research is engaged in determining the accuracy and repeatability of the test (Lancet, 1974), and also its sensitivity and specificity. So far it has been established that, as well as detecting 'open' spina bifida, the test detects anencephaly (probably more consistently (Lancet, 1974)). In addition, a raised serum AFP may be associated with a normal twin pregnancy, exomphalos, or intrauterine death. Cases of 'closed' spina bifida are not detected.

A definitive diagnostic procedure should be available for the confirmation, or otherwise, of positive screening results. In screening for spina bifida, twins and intrauterine death would be excluded by ultrasonography, and normal pregnancy by measurement of amniotic fluid AFP. Both procedures are of proven diagnostic efficacy; more particularly, the risk of mis-diagnosis and termination of a normal pregnancy is negligible, and a recent report (Bartsch, Lundberg, and Wahlstrom, 1974) suggests that the risk of spontaneous abortion after amniocentesis is very low. Raised amniotic fluid AFP levels are, however, associated with anencephaly and exomphalos as well as 'open' spina bifida; this would not however, raise practical difficulties.

Effective and acceptable treatment should be available to those in whom the diagnosis is confirmed. In screening for spina bifida, pregnancies found to be affected with 'open' spina bifida, anencephaly, or exomphalos would be terminated. In practice this has been found acceptable to women who have previously had an affected child,

although no population-based study has been reported. Its acceptability under conditions of mass-screening is obviously still unknown.

The costs of establishing and running a screening service should be set out, together with the economic benefits to be derived from its introduction. It is the purpose of this paper to examine whether the considerable investment that would be involved in the establishment and maintenance of a mass-screening programme could be justified in terms of the economic benefits to be derived from reducing the number of children born with 'open' spina bifida.

METHOD

The study was based on statistics of the Western Regional Hospital Board (WRHB), Scotland, area in 1973.

Estimates were made of the number of children born with spina bifida cystica in the WRHB area in 1973, and of the probable number of survivors and their degree of handicap. Survivors with spina bifida use more resources—medical, educational, social, and personal—than those without handicap. An estimate of the average excess use of resources was calculated.

The characteristics of a typical mass-screening programme for spina bifida were set out. The total costs to the Health Service and to the patients of running such a programme for 20 years were calculated. The annual number of births of affected children which its introduction would prevent was derived. Thence, the saving of excess costs in resource use, through births prevented as a result of a 20-year screening programme was calculated. This represented the economic benefits of such a programme.

All prices were standardized to July 1974 levels using the Retail Price Index (Information Division of the Treasury, 1974). Discounting was employed throughout to obtain net present values of costs and economic benefits. The net present value of the economic benefits was divided by that of total costs to produce a benefit-cost index. The practical usefulness of the benefit-cost index was examined.

FINDINGS

EPIDEMIOLOGY AND NATURAL HISTORY

NUMBER OF PREGNANCIES AFFECTED In 1973 there were 43 594 births in the WRHB area. Calculations were based on the screening of 43 000 pregnancies per annum. In a study of births in Glasgow during the years 1964-68, Wilson (1970)

found the incidence of spina bifida cystica to be 2·80/1000 total births; a similar incidence (2·83/1000) was recorded for anencephaly. Findings of 2·94/1000 (spina bifida) and 2·78/1000 (anencephaly) were recorded by the Glasgow Congenital Malformations Registry during the years 1972-73. No such data are available for the other counties of the WRHB. However, published statistics for spina bifida and anencephalic stillbirths (Registrar General for Scotland, 1968-72) show no significant difference in the incidence of either condition between Glasgow and the rest of the WRHB area. The incidence rates at birth of each condition recorded in Glasgow since 1964 (2·8/1000 total births for each) were therefore taken as the best available estimates of the incidence rates at birth throughout the WRHB area in 1973. It was assumed that the prevalence rates of affected fetuses at the time of screening were equal to the estimated birth incidence rates. Such prevalence rates would be associated with 120 of 43 000 pregnancies being affected with spina bifida cystica and a further 120 with anencephaly.

OUTCOME OF PREGNANCY Most anencephalics are stillborn and the rest die within a few days. Since their use of resources is therefore negligible these were not considered. Of 120 pregnancies affected with spina bifida, on average 114 would be expected to result in births with myeloele, and six with meningocele (Laurence and Weeks, 1971). The resources used by those with meningocele were not considered, since almost all have closed lesions and their presence would not be detected by the proposed screening test; in any case, most of them have little handicap. Of 114 children born with myeloele, an average of 92 live births and 22 stillbirths would be expected (Richards and McIntosh, 1973). Current experience in the west of Scotland suggests that, in the absence of a specific campaign, a further two births may be prevented through the uptake of amniocentesis and selective termination by 'higher-risk' women. This would result in a reduction of births with spina bifida to 118, of which 90 would be liveborn with myeloele and six with meningocele.

SURVIVAL From published statistics on spina bifida stillbirths during the period 1968-72, for the WRHB area and for Scotland (Registrar General for Scotland, 1968-72), an estimated national incidence for spina bifida of 2·3/1000 total births was derived. Using infant mortality statistics for the same period, the survival rate of all spina bifida births at one year was estimated to be 47%. A rate of 47%

surviving infancy, recorded by the Glasgow Congenital Malformations Registry for the years 1972-73, accords with the national picture.

Stark and Drummond (1973), however, reported a lower survival rate in a series in which those born with myeloele in 1965-71 were carefully selected for early surgery, and the equivalent of only 38% of all spina bifida births survived infancy. The potential results calculated, and early results reported, of such a policy, by Lorber (1971, 1972, 1973) pointed to an even lower survival rate. A likely explanation of the recent high survival rate in Scotland is that selective surgical policies had not then been fully introduced.

For the purposes of this study it was assumed that, with fuller implementation of careful selection for early surgery, and continuing improvement in techniques, the survival of all spina bifida births at one year would be 45%, that is 53 of 118. A variety of reports show that a further 5% to 7% die before they reach the age of five years (Stark and Drummond, 1973; Laurence, 1974). Survival of all spina bifida births at five years of age was assumed to be 40%, that is 47 of 118. Assuming that all six children with meningocele survived to the age of five years, the number of children with myeloele surviving infancy would be 47, of whom 41 would reach their fifth birthday.

HANDICAP There have been numerous reports of degree of handicap among survivors (Lorber, 1971; Laurence, 1974; Wilson, 1971; Smith and Smith 1973). Stark and Drummond (1973) reported a series of reasonable size, gathered over a number of years, on whom a selective surgical policy was applied. Since it is reported that selective policies are widely accepted, it is thus more appropriate to use their findings than those for universally operated series, totally unoperated series, or those in which the presumptive outcome of the application of selective policies has been calculated. Applying Stark and Drummond's findings, and using the categories defined by Lorber (1971), the degree of handicap at five years among survivors with myeloele was estimated (Table I).

LIFE EXPECTANCY For the purposes of this study it was assumed that those in handicap category 2 had the same life expectancy as the general population of Scotland at five years of age (Registrar General for Scotland, 1968-72). There are no firm data on the mortality of those in other categories. However, reports of potential morbidity and early mortality from a variety of complaints

TABLE I
DEGREE OF HANDICAP AT FIVE YEARS

Category	Physical Handicap	Mental Capacity	Survivors (%)
2 ..	Moderate	Normal intellect	25
3 ..	Severe	Normal intellect	55
4 ..	Severe	Moderate mental retardation	13
5 ..	Severe	Severe mental retardation	7

made the adoption of a mortality curve following a cosine wave pattern seem appropriate. This is illustrated in Fig. 1.

FIG. 1. Assumed survival curve for severely handicapped myelocoele sufferers (categories 3, 4, and 5).

SERVICE PROVISION: EXCESS USE OF RESOURCES BY THOSE LIVEBORN WITH MYELOCELE

The use of resources by 90 children liveborn with myelocoele in excess of those used by 90 normal children is considered under the headings hospital treatment, permanent care, education, loss of maternal income, and additional costs. An estimate of the excess costs which would be incurred in a number of years after birth is presented under these headings which are further subdivided as shown in Table II. The derivation of these costs is indicated below. Detailed consideration of the costing is beyond the scope of this paper, but is available elsewhere (Haggard, Carter, and Milne, 1975).

HOSPITAL TREATMENT

INPATIENT Data were obtained on request from the Information Services Division of the Scottish Health Service and the accounts of the WRHB

for 1974. The average time spent in hospital by survivors with myelocoele fell with age, from 44 days per annum by children aged less than one year, for example, to eight days per annum at five years of age, and less in later childhood. Average costs per child inpatient day, including an estimate for visitors' travelling costs, were found to be just under £23. Total annual costs of providing inpatient care for all myelocoele survivors at various ages are set out in Table II. These costs fell steeply from about £90 000 for infants, to about £10 000 in early childhood, and £5000 or less in later childhood.

OUTPATIENT Data were obtained from a study of spina bifida children (Zealley, 1974), a study of ambulance costs (Davidson, 1974), and the accounts of the WRHB for 1974. Outpatient visits, averaging two during the first year of life and eight per annum thereafter, were found to cost a little over £4 per visit, including travelling. Table II shows that the total annual cost of outpatient visits was about £400 in infancy and £1300-£1400 throughout most of childhood.

PHYSIOTHERAPY Physiotherapy is chiefly required in childhood. No population-based data are available. An assumption was made that at any time a quarter of children of between 3 and 10 years of age receive fortnightly physiotherapy which costs the same as an average outpatient visit. Table II shows that these assumptions led to an estimated annual total cost for physiotherapy of about £1100.

PERMANENT CARE There are always likely to be some affected children needing to enter permanent care because their families can no longer cope (Hunt, 1973; Lorber, 1972). No population-based data are available. It was assumed that 5% of those surviving entered permanent care at the age of five years, that 10% of all survivors were in permanent care at 16, and 15% at 30 years, and that all entrants to permanent care were seriously handicapped (categories 3-5). The cost taken to be representative of permanent care of whatever kind, was that for a patient in a WRHB mental deficiency hospital—£1293 per annum in 1973-74. From this figure the saving to a family through the admission of a child to permanent care was subtracted—£254 at 1972 prices (Department of Employment, 1972). For adults in permanent care an estimate of their total consumption expenditure was subtracted—£1040 at 1973 prices (UK Central Statistical Office, 1974). The total annual cost of providing permanent care, shown in Table II, amounts to £2290 in childhood and rather less thereafter.

TABLE II
COSTS OF CARING FOR 90 SURVIVORS WITH MYELOCELE

Year	No. of Survivors			Costs							Additional Costs		Total Cost	Present Values Discounted at		
	Handicap Category		Total	Hospital Treatment			Perma- nent Care	Educa- tion	Maternal Income							
	2	3-5		In- patient	Out- patient	Physio- therapy										
	£	£	£	£	£	£	£	£	£	£	£					
0	—	—	90	89 930	390	—	—	—	5550	1850	—	97 720	97 720	97 720	97 720	
1	—	—	47	6400	1480	—	—	—	5550	1850	—	15 280	14 550	13 890	13 290	
2	—	—	45	13 290	1410	—	—	—	5550	1750	—	22 000	19 950	18 180	16 630	
3	—	—	43	14 650	1380	1180	—	2850	4440	3300	—	27 800	24 010	20 890	18 280	
4	—	—	42	8580	1350	1180	—	2850	4440	3200	—	21 600	17 770	14 750	12 350	
5	10	31	41	7450	1350	1070	2290	11 040	7770	3100	—	34 070	26 690	21 150	16 940	
12	10	30	40	3450	1000	—	2290	6970	9990	3000	—1220	25 480	14 190	8120	4760	
16	10	29	39	2020	—	—	1010	—	—	—	7910	10 940	5010	2380	1170	
21	10	26	36	—	—	—	1010	—	—	—	28 140	29 150	10 460	3940	1550	
30	10	20	30	—	—	—	1260	—	—	—	27 890	29 150	6750	1670	—	
40	10	11	21	—	—	—	500	—	—	—	27 360	27 860	3960	620	—	
55	9	0	9	—	—	—	—	—	—	—	26 200	26 200	1790	140	—	
Lifetime totals:												1 059 000	573 600	354 800	267 700	

EDUCATION

NURSERY Local authorities provide extra nursery places for children with spina bifida. No population based data are available, although a survey in south-east Scotland (Woodburn, 1974) revealed that 70% of three to four-year-olds with spina bifida were attending nursery schools. This provision was assumed for the WRHB area; it compares with an intended provision for 45% of normal three and four-year-olds. The annual cost per pupil in nursery schools in Glasgow during 1973-74 was £236. The total annual excess cost of nursery education was found to amount to about £3000 (see Table II).

PRIMARY, SECONDARY, AND OTHER From estimates of distribution of handicap, and available data on current educational provision (Woodburn, 1974; Hamilton, 1974), the likely requirements for different forms of education among the 95% of survivors with myelocoele not admitted to permanent care at the age of five years, were estimated to be:

35% able to attend normal schools, 50% at special day schools, 5% at special residential schools, and 5% at Junior Occupational Centres. The cost in Glasgow during 1973-74 of special schools was £382 a pupil per annum, and of Junior Occupational Centres £502 (City Chamberlain, Glasgow, 1974). In 1974 the cost in Scotland of residential special schooling for the physically handicapped was £2250 per pupil per annum (Scottish Education Department, 1974). To the costs of non-residential provision was added an estimate for average excess travelling costs (assumed to be £40 per annum). From all special education costs were subtracted the costs of ordinary schooling at the appropriate ages—£116 a pupil per annum in a primary school in Scotland during 1971-72, and £234 in a secondary school (Scottish Education Department, 1972). The total excess costs of providing special education were found to amount to £11 000 per annum in the primary years and £7000 per annum at the secondary stage (see Table II).

LOSS OF MATERNAL INCOME

Some mothers with handicapped children are prevented from going out to work unless they employ someone else to care for the child while they are away. In the absence of other data it was assumed that 70% of the mothers of seriously handicapped children who might otherwise have gone out to work were prevented from so doing. In 1972 the percentage of mothers employed varied from 20 to 50% depending on the ages of dependent children, and their average weekly earnings were £16.50 (UK Central Statistical Office, 1973). From these statistics an estimate of the total annual cost of lost maternal income could be calculated. As shown in Table II, it rises from about £5000 in early childhood to £10 000 in later years.

ADDITIONAL COSTS

IN CHILDHOOD Families with handicapped children incur extra expenses in clothing and equipment. A number of families move house because of the handicapped child. They make greater use of health and social services—such as, health visitors and social workers—and many children need orthopaedic appliances. Although no population-based data are available, preliminary data supplied by the recently established Family Fund give some indication of the scale of personal requirements and their costs. It has been assumed that average extra family expenditure for seriously handicapped children of between three and 15 years of age is £50 per annum, and that further expenditure of £50 per annum is incurred through the extra provision of non-hospital health and social services to seriously handicapped children from birth to 15 years. Total additional costs in childhood amount to about £2000-£3000 per annum (see Table II).

OF DEPENDENT ADULTS The severity of handicap affects a survivor's ability to participate in gainful employment. No population-based data on employment of survivors with myelocoele exist. Taking into account the findings of a recent study (Evans, Hickman, and Carter, 1974), it was assumed that 66% of adult survivors would enjoy full-time employment, 12% would often be unemployed, and 22% would never be employed. The excess lifetime costs to society of adults often or always unemployed are their total lost earnings, less the value of any consumption expenditure not incurred in the event of premature death. As can be seen from Table II, the total annual cost of dependent adults is about £8000 at 16 years of age and almost £30 000 for the greater part of adult life (ages 21 to 55 years).

TOTALS

The total estimated excess annual costs incurred by all survivors with myelocoele for each age group are obtained by adding together the estimate under each column heading; they are shown in the 'total' column of Table II and can be seen to amount to almost £100 000 in the first year of life and to vary thereafter between about £10 000 and £35 000 until 55 years of age. The addition of the totals relating to each year of life gives a grand total of £1 059 000 as an *apparent* measure of the excess lifetime cost resulting from the birth of 90 live children with myelocoele. However, such an addition of expenditures valued at today's prices, but expected over a number of years into the future, is not acceptable as a valid total expenditure during the period in question. This is because a pound saved in one year's time would not be as valuable as a pound saved now, for the present pound could be loaned or invested at a positive rate of interest, producing something more than a pound a year from now. And in order to have a pound saved in one year's time it is necessary to set aside less than a pound now—still less to obtain a pound in two, three, or more years in the future. Thus, expressing anticipated future costs at current prices overstates their present values. To overcome this difficulty, a rate of discount is normally applied to the current prices of anticipated future costs in order to obtain the (lower) present values. For reasons which are set out elsewhere (Hagard *et al.*, 1975), a discount rate of 10% (currently employed by the UK Treasury) has been taken as the most appropriate for use in this study. For comparison, the effects of discounting at 5% or 15% have also been shown in Table II, and present values of annual totals and lifetime totals after discounting are given in the three right-hand columns. The present values of total excess lifetime costs resulting from the birth of 90 live children with myelocoele in the WRHB area, discounted at 5, 10, and 15% were found to be £573 600, £354 800, and £267 700 respectively, and the present values of the respective excess costs relating to one such child £6370, £3940, and £2970.

CHARACTERISTICS OF A SCREENING PROGRAMME FOR SPINA BIFIDA

Calculations were based on 43 000 women at risk of having 120 children with spina bifida and 120 with anencephaly, and are summarized in the flow chart (Fig. 2).

Of the fetuses affected with spina bifida approximately 15% (18) would have had 'closed' lesions

FIG. 2. Flow chart illustrating the characteristics of a screening programme for spina bifida

(Laurence, 1974); these would not have been detected and would, therefore, be associated with the births of affected children. Thus 120 cases of anencephaly, but only 102 of spina bifida, would have been available for detection.

ATTENDANCE It was assumed that 90% (38 700) of women would attend antenatal clinic at the appropriate time for screening (15th-20th week of gestation), that attendance could be so arranged that this would entail no extra visits, and that the characteristics of attenders and non-attenders would be the same. It was further assumed that all attenders would participate fully in the programme.

Attenders would have 90% of 102 (92) pregnancies affected with 'open' spina bifida, and 90% of 120 (108) pregnancies affected with anencephaly; thus non-attenders would give birth to 10 children affected with 'open' spina bifida, and 12 with anencephaly.

HIGHER RISK PREGNANCIES These were assumed to derive from ongoing totals of 120 spina bifida and 120 anencephalic births annually, and were estimated to amount to 333 pregnancies per annum (Hagard *et al.*, 1975). At an attendance rate of 90%, 300 of 333 women at higher risk would be found among the 38 700 attenders. At a genetic counselling session these would be offered direct diagnosis (by amniocentesis) without screening. Taking the overall incidence of neural tube malformation in higher risk pregnancies to be 10 times the normal population incidence, the presence of seven fetuses with 'open' spina bifida and eight with anencephaly would be diagnosed, and the 15 pregnancies terminated. This would leave, among the 38 400 pregnancies remaining to be screened 92 minus seven (85) with 'open' spina bifida, and 108 minus eight (100) with anencephaly; 38 215 would not be affected with either malformation.

TEST CHARACTERISTICS Since serum AFP rises as pregnancy proceeds, gestational age needs to be accurately known. When this is uncertain it can be measured by ultrasonography. On the basis of a recent study (Beazley and Underhill, 1971), it was assumed that a fifth of those screened (7680) would need ultrasonographic dating, necessitating an extra attendance at the clinic. On the basis of experience in the west of Scotland, it was assumed that as many as 10% (3840) might require a second serum AFP estimation; this would usually also necessitate an extra attendance at the clinic.

A diagnostic procedure would be required for all those screening positive. In almost all cases this would entail amniocentesis. The number of false

positives would need to be kept as low as possible, both 'to avoid needless anxiety, and to prevent iatrogenic disease associated with the diagnostic process itself' (Klarman, 1974); to achieve this a high test specificity would be required. A test specificity of 99.5% would be associated with 38 024/38 215 (99.5%) of unaffected pregnancies screening true-negative and the remainder (191) screening false-positive. This test specificity was assumed. The setting of a very high test specificity would be associated with considerable limitation in the sensitivity of the test (Cochrane and Holland, 1971), that is in its ability to detect true-positives. A test sensitivity of 80% might be possible with a specificity of 99.5%*. This sensitivity was assumed; it would be associated with 68/85 (80%) of pregnancies affected with 'open' spina bifida screening true-positive and 17/85 screening false-negative, and with 80/100 affected with anencephaly screening true-positive and 20/100 false-negative. Thus a further 17 cases of 'open' spina bifida and 20 of anencephaly would not be detected and would be associated with the births of affected children.

All cases screening serum-positive would require diagnosis; these would comprise 191 screening false-positive, and 68 'open' spina bifida and 80 anencephaly screening true-positive, giving a total of 339. Approximately four additional serum positives would be found associated with cases of twin pregnancy, intrauterine death (rarely), with exomphalos (very rarely).

DIAGNOSIS All 343 women screening serum-positive would have a further ultrasonographic examination, and those (339) in whom twins or intrauterine death were not detected would receive amniocentesis. Diagnosis of neural tube malformation would be confirmed in 68 cases with 'open' spina bifida and 80 with anencephaly; the 148 pregnancies would be terminated.

OUTCOME Altogether, a screening programme as described would result in the detection and termination of 75 of 120 pregnancies affected with spina bifida (and 88/120 affected with anencephaly).

SERVICE PROVISION: COSTS OF ESTABLISHING AND MAINTAINING SCREENING FOR SPINA BIFIDA

The costs of providing screening for spina bifida are considered under the headings shown in Table III. Derivation of these cost estimates is

* A test sensitivity of 80% was widely expected at the time the paper was received for publication (April 1975). Subsequent evaluation in many centres suggests that a sensitivity for myelocoele of 40 to 50% is more likely. The economic consequences of lower test sensitivity are considered in the discussion.

TABLE III
COSTS OF THE SCREENING PROGRAMME

Service Provision	Costs					
	Capital				Recurrent	
	Recurring after (years)				To Health Service	To Patients
	3	5	10	20 or more	£	£
Publicity	£	£	£	£	20 000	
Genetic counselling (539 consultations) Staff (≡ 6 senior registrar sessions/week)					2513	3126
Patient at £1.60 (women) and £4.20 (men)						
Amniocentesis (539 examinations) Staff (≡ 4 registrar sessions/week)					1448	2695
Patient at £5.00						
Ultrasonography (8023 examinations) Accommodation				24 100		
Equipment			10 000			
Staff (≡ 3 full-time midwifery sisters) personnel training			1154		6136	
Patient at £1.60						12 837
Extra clinic attendance (3840 attendances) Staff (≡ 3 registrar and 3 staff midwife sessions per week) ..					1571	6144
Patient at £1.60						
Radioimmune assay (42 779 AFP estimations) Accommodation				11 190		
Equipment	5500	15 500	680		10 400	
Laboratory staff					1138	
materials					44 020	
Reagent and anti-serum					1473	
Dispatch						
Column totals:	5500	15 500	11 834	35 290	88 699	24 802
Total				68 120		113 500

considered below. Detailed consideration of the costing is beyond the scope of this paper but is available elsewhere (Hagard *et al.*, 1975).

PUBLICITY A publicity campaign aimed at achieving 90% uptake of screening would need to be directed at professional health workers, particularly obstetricians and general practitioners, and at the 'at-risk' group itself. On the basis of data supplied by the Scottish Health Education Unit an expenditure of £20 000 per annum for such a campaign has been assumed.

GENETIC COUNSELLING AND AMNIOCENTESIS On the basis of current experience in the west of Scotland, it was estimated that demand for genetic counselling and diagnostic amniocentesis among women at higher risk would reach 100 per year if no screening service were introduced, and that introduction of such a service would thus result in an extra 200 such consultations and examinations annually. It is the costs which these

would generate that have to be considered, together with the costs resulting from provision for 339 women screening serum-positive. They would consist of recurrent costs to the Health Service through the provision of salaries, and to patients and their husbands through lost earnings and travelling expenses. Health Service costs were calculated at £2513 per annum for genetic counselling and £1448 per annum for amniocentesis, while the respective costs to patients were found to be £3126 and £2695 (see Table III).

ULTRASONOGRAPHY A total of 8023 ultrasonographic examinations would be required—7680 for dating and a further 343 for those screening serum-positive. This would entail capital costs to the Health Service for the provision of accommodation, equipment, and personnel training, recurrent costs to the Health Service for salaries, and costs to the patients through lost earnings and travelling expenses. Capital costs to the Health Service were estimated to be £35 254; every 10 years, on average,

equipment replacement (costing £10 000), and additional staff training (costing £1154) would be required. Recurrent costs to the Health Service were estimated at £6136 per annum and to the patients at £12 837 per annum (see Table III).

EXTRA CLINIC ATTENDANCE Altogether 3840 extra clinic attendances for a second blood sample to be taken would entail recurrent costs to the Health Service in provision of staff (£1571 per annum) and for materials and dispatch (considered under 'radioimmune assay'), and to the patients (£6144 per annum) (see Table III).

RADIOIMMUNE ASSAY A total of 38 400 initial serum samples, 3840 repeat serum samples, and 539 amniotic fluid samples would generate 42 779 AFP estimations. This would entail capital costs to the Health Service for accommodation (estimated at £11 190) and equipment (estimated at £5500 for that having a three-year life, £15 500 for that with a five-year life, and £680 for that needing to be replaced after 10 years' use), and recurrent costs to the Health Service for staff (estimated at £10 400 per annum), materials (£1138 per annum), reagent and anti-serum (£44 020 per annum), and dispatch (£1473 per annum). No cost to patients would be incurred (see Table III).

TOTAL COSTS OF SCREENING

The costs of running a screening programme must be seen in terms of initial capital costs, recurrent costs (for example, staff), and interval replacement (for example, equipment as it ceases to be of use). The capital costs of establishing the programme would be borne by the Health Service and amount to a total of £68 120. Recurrent costs to the Health Service would be £88 699, and to the patients £24 802. These costs, totalling £113 500, would recur in each year of the programme. Replacement costs, amounting to £5500, would recur every three years, to £15 500 every five years, and to £11 834 every 10 years (see Table III). Accommodation would not require replacement within the first 20 years of the programme.

Using these data it is possible to calculate the costs which would be incurred in the first, second, and third year of the programme, and so forth. For the purposes of this study, the costs of running a screening programme for 20 years were calculated.

The addition of:

Initial capital costs amounting to	£68 120
Three, five, and 10 year interval replacement costs totalling ..	£91 330
And 20 years' recurrent costs at £113 500 per annum ..	£2 270 000
gives a total of	£2 429 450

for such a programme. However, as already argued, the present *value* of costs to be incurred in the future would be less according to how far into the future they would be incurred. Therefore, discounting at an appropriate rate is required to obtain the net present values of costs expected in each of the 20 years.

This procedure was employed in precisely the same manner used in calculating benefits (illustrated in Table II), and the present values of the total costs which would be incurred in establishing and running a 20-year screening programme were found to be:

£1 610 070 using a 5% rate of discount
£1 168 530 using a 10% rate of discount
£911 200 using a 15% rate of discount.

ECONOMIC BENEFITS OF THE SCREENING PROGRAMME

It has been shown that in the absence of screening, the likely outcome of 120 pregnancies affected with spina bifida in the WRHB area each year would be:

Terminations	2
Stillborn	22
Liveborn with myelocoele ..	90
Liveborn with meningocele ..	6

and that the introduction of the screening programme described would result in 75 terminations. Of the 45 affected pregnancies not terminated, six would be affected with meningocele. The other 39 would result in children being born with myelocoele, of whom about 20% (eight) would be stillborn, and the remainder (31) liveborn. Thus, the screening programme would result in the prevention of the birth of 90 minus 31 (59) liveborn children with myelocoele each year.

The economic benefits of preventing the birth of one liveborn child with myelocoele have been calculated at:

£6370 where a 5% discount rate is employed
£3940 where a 10% discount rate is employed
£2970 where a 15% discount rate is employed
(see Service Provision, page 43).

Thus, the economic benefits to be derived from running the screening programme for one year would be:

£6370 × 59 (£375 830) using a 5% discount rate

£3940 × 59 (£232 460) using a 10% discount rate

£2970 × 59 (£175 230) using a 15% discount rate.

Therefore, if the screening programme were run for 20 years these annual economic benefits could be expected in each year. However, since the net present value of such benefits would be less according to how far into the future they were obtained, discounting at the appropriate rate would again be required to obtain the net present values of benefits for each of the 20 years. The sum of these net present values would represent the net present value of total economic benefits of running the screening programme for 20 years. It was calculated, and found to be:

£4 920 200 using a 5% discount rate

£2 178 090 using a 10% discount rate

£1 263 360 using a 15% discount rate.

THE BENEFIT-COST INDEX

This was derived by dividing the total economic benefits which the screening programme would produce, by its total costs.

That is:

£4 920 200 by £1 610 070 to give 3.06 using a 5% discount rate

£2 178 090 by £1 168 530 to give 1.86 using a 10% discount rate

£1 263 360 by £911 200 to give 1.39 using a 15% discount rate.

As already stated, discounting at 10% was felt to be most appropriate to this study. Therefore, the derivation of a Benefit-Cost Index (BCI) of 1.86, that is, of economic benefits outweighing costs in the ratio 1.86 : 1, is the main finding of the exercise.

DISCUSSION

While studies of this kind have to be based on the epidemiology and natural history of a disease in a defined area of an appropriate size, the nature of the technological procedures involved requires that their provision is considered only in relation to large centres. The geography and pattern of medical services in the west of Scotland is such that if a programme of mass-screening in pregnancy for spina bifida were to be introduced in the region, it would be offered to the population served by the six Health Boards which have provided all medical services since 1 April 1974, and would

involve the radioimmune assay facilities available only in Glasgow. Since the only useful regional statistics available for this study related to the WRHB area, the territory for which it was responsible until 31 March 1974 was taken as the defined area for the exercise. In practice it scarcely differs from that of the six successor Health Boards.

If this kind of study is to be feasible, reliable data relating to the epidemiology and natural history of the disease for which screening is proposed ought to be available and, moreover, in a form which can be linked to adequate data of resource utilization and costs. Had hospital statistics been available in patient- rather than incident-related form (Heasman, 1968), and had evaluations of their work been available from clinical divisions, many of the assumptions made in this paper would have rested on firmer foundations. Similar considerations apply to the usefulness of education and social work statistics. Nevertheless, the epidemiology was well enough known to provide a more reliable basis than is often the case in studies of this nature (Klarman, 1965), and the BCI of 1.86 can be regarded as giving a good indication of the relationship between costs and benefits in the particular situation defined. The BCI would, however, be sensitive to changes in methodology and input assumptions. Those which would have the greatest effect on its value—such as, the use of a different discount rate, alternative treatment policies, lower antenatal attendance, refusal of amniocentesis, 'free' ultrasoundography, and cheaper reagent—are discussed.

DISCOUNT RATE While economists are agreed that it is necessary to employ a discount rate, they do not agree on its size. It is beyond the scope of this paper to consider the merits of choosing between different discount rates. The use of a 5% rate would have the effect of raising the BCI to 3.06, that of a 15% rate of lowering it to 1.39.

TREATMENT POLICIES The calculations rest on two controversial aspects of current medical practice. In the first place the termination of pregnancies affected with severe fetal malformation is lawful and acceptable, whereas euthanasia (of affected neonates) is not. If it were, this exercise would be superfluous; the cost of screening might, therefore, be regarded as payment for a scruple. Secondly, current selective surgical policy towards affected neonates involves assessing both the likelihood of survival without operation and

the probable extent of eventual handicap if operated, and then not operating on those for whom only surgical intervention would be likely to secure survival, but who would nevertheless have little prospect of avoiding severe handicap. A less selective surgical policy, such as that in general currency until recently, produces more survivors, with a greater burden of handicap. In the context of this study, it would therefore be associated with a higher value for the BCI. A more rigorous selective approach would result in fewer survivors, probably with less handicap, and the screening programme would thus produce a smaller economic benefit. On the other hand, since less than ideal provision is currently available to survivors with spina bifida, were there fewer survivors more resources could be devoted to each; the prevention of additional births would thus result in greater resource savings and help to raise the value of the BCI. Fewer survivors would, of course, result not only from the introduction of a more rigorous selective surgical policy but also through the provision of screening itself.

ANTENATAL ATTENDANCE All the capital costs of the screening programme, and a certain amount of the recurrent costs, would need to be committed whatever antenatal attendance eventually materialized, whereas benefits would be higher or lower in proportion to attendance. The effect on the BCI of a 50% (21 500) attendance, rather than 90% (38 400) would be to diminish its value from 1.86 to 1.44. Furthermore, considerable differences in the characteristics of attenders and non-attenders might be expected. Women in social classes IV and V probably have a higher incidence of affected pregnancies (Edwards, 1958; Wilson, 1971), and are known to be less likely to attend at the appropriate time for screening (McKinlay, 1970). Recognition of this would be required in planning the publicity campaign, although experience of both conducting and evaluating campaigns aimed at particular target groups has so far been very limited (Jones and Grahame, 1974).

REFUSAL OF AMNIOCENTESIS This would also help to lower the value of the BCI. Almost all costs, except those for amniocentesis itself, would have been incurred, and no economic benefits would be expected to flow from them. Experience in Glasgow suggests that only a tiny percentage of pregnant women at *higher* risk would not wish amniocentesis and termination of affected pregnancies. Of 339 women screening serum-positive, on average 68 would be carrying fetuses

affected with spina bifida, 81% (55) of whom would be liveborn were pregnancy to continue to term. While predictions of the likely behaviour of women screening serum-positive must be undertaken with caution, the findings of a recent survey suggested that as many as 70% of people in the UK approve abortion for a pregnant woman who knows she may have a severely deformed baby (Action Research for the Crippled Child, 1974). It is reasonable to assume that an even higher percentage of women actually screening serum-positive would request amniocentesis and selective termination. In an area such as the WRHB this may not be true, since a greater than average proportion of the population is of Roman Catholic persuasion. Refusal of amniocentesis and selective termination by 30% (102) of those screening positive would lower the BCI from 1.86 to 1.37.

'FREE' ULTRASONOGRAPHY AND CHEAPER REAGENT Two developments in screening may be anticipated which would help to decrease the recurrent costs of screening. Ultrasonographic examination is likely to become a routine procedure during the second trimester of pregnancy (Chard, 1974). The removal of the cost of its provision from the screening service would help to increase the value of the BCI from 1.86 to 2.29. The introduction of mass-serum screening would be likely to lead to the development of less expensive radioimmune assay reagents and technique. A 50% reduction in reagent costs would help to increase the BCI from 1.86 to 2.26, and a 75% reduction to increase it from 1.86 to 2.53.

From the foregoing the main conclusion to emerge from this study is that the economic case for establishing a screening programme for spina bifida in the west of Scotland is strong. Now, assuming broadly similar costs and comparable economies of scale in areas where the incidence of spina bifida is greater or less than 2.8/1000 total births, the BCI would be respectively higher or lower, and the economic case for introducing screening correspondingly stronger or weaker. Similarly, failure to develop a screening test with sensitivity as high as 80% would lead to a lower BCI and a weaker economic case for introducing screening.

Fig. 3, which relates the sensitivity of the screening test with the birth incidence of the condition, demonstrates the practical importance of this exercise. At a test sensitivity of 80%, the value of the BCI would be above one in all regions of the UK for which reliable surveys of spina

FIG. 3. Variation of benefit-cost index showing screening test sensitivity and incidence of spina bifida

bifida birth incidence exist. But if test sensitivity were only 20%, the value of the BCI would not exceed unity in any region. The consequences of health service planning decisions in this, as in most fields, affect society as a whole, and those who take them must perceive them beyond the narrow framework of health service administration and accountability. Implicit in such a perception is the acceptance of cost-benefit analysis, which is concerned with whether society as a whole is made economically better off by undertaking, rather than by not undertaking, a particular project. If, for example, the test sensitivity were finally shown to be 40%, those responsible for health service planning in Belfast, where the incidence of spina bifida was 3.6/1000 total births (Elwood, 1972), would have economic support for a decision to introduce screening, even if to the Health Service alone the costs considerably outweighed the economic benefits. In Birmingham, where the spina bifida incidence is 2.0/1000 total births (Knox, 1967), a test sensitivity of 40% would be associated with a BCI of about 0.7. Such an eventuality might, but should not, be interpreted as implying that no case would exist for introducing screening in Birmingham or, indeed, that a case might be developed if net profit to the Health Service could be shown. It is not a legitimate objective in this kind of exercise to maximize the BCI unless in association with the same or greater community benefits. On the other hand, the social and political implications of not screening for spina bifida in Birmingham might well be such that the health authority would be prepared to accept the economic costs involved. The value of

cost-benefit analysis in the preparation and execution of that kind of health service planning decision lies in the improvement in sophistication which it brings to the process.

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