Contemporary Themes

Meningomyelocele: The Price of Treatment

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This study was originally carried out to compare the management of patients with meningomyelocele at the London Hospital with that at Sheffield Children's Hospital (where a particular clinical interest in this problem exists) with a view to improving the arrangements at the former. It soon became apparent that to set up a service along the lines of that in Sheffield would be extremely expensive and would place immense demands on the diagnostic and therapeutic facilities of a hospital whose resources were already stretched to the maximum. It was therefore decided to assess, if possible, the cost to the taxpayer of a nationwide service in England and Wales. Many of these calculations are nothing more than educated estimates, but they do serve to show at least the minimum cost.

In the last decade of the nineteenth century 153 babies in every 1,000 live births died in the first year of life. In 1965 this figure had been reduced to 19 deaths per 1,000 live births.[1 2] Of these 19 deaths 21% were due to congenital abnormalities, and 0·5 per 1,000 of these were meningomyeloceles. Until about 14 years ago 94% of infants with a meningomyelocele died early of meningitis, progressive hydrocephalus, or the complications of paraplegia. Of the remainder 4% lived as permanent invalids in chairs and 2% managed some form of life after multiple operations.[1] Thus absence of treatment brought with it the inheritance of survivors whose mental and physical abilities left much to be desired, but who represented only 6% of affected children. These figures were produced by a policy of treatment which would now be regarded as treatment by neglect. An ethically acceptable minimum of treatment now produces a 30% survival rate at three years.[3] Many of these survivors will be severely disabled, and about half will need almost permanent institutional care. The remainder all need special educational facilities and a great deal of surgical treatment.

The use of antibiotics, bypass procedures for the hydrocephalus (which occurs in about half of these patients), and closure of the spinal defect within the first 24 hours of life have greatly improved the expectation of life. At 2 years 70% of these patients will be alive and 40% will survive to adult life, a good proportion achieving some independence.[1]

Figures based on the experience of the Sheffield Children's Hospital serve to indicate the size of the resulting therapeutic problem.[2] These figures are for all patients with spina bifida cystica, and are therefore optimistic. Nevertheless, as 86·5% of such patients in Sharrard's study[3] had meningomyeloceles these figures are a reasonable parallel, though not totally accurate. For every 100 patients 30 die. Of the remainder 15 have no urinary complications, 25 require operation resulting in satisfactory urinary function, 25 have satisfactory urinary function with conservative management, and 5 die of urological complications. Twenty-two will require limited or no orthopaedic procedures, 27 will require multiple operations to obtain good mobility, 13 will require multiple operations to obtain partial independence, and 8 will require wheel-chairs. Fifty-five will be mentally normal but physically handicapped, 10 will require education at special schools, and 5 will be ineducable.

The resulting educational problems were discussed by McKeown,[4] who quoted figures obtained by Knox[5] when the latter investigated the consequences of medical measures in this condition. In the 65,935 births in Birmingham in the years 1960-2 there were 132 infants (live-born or stillborn) with spina bifida. At four to five years after birth 13 were alive and well and 14 were alive but disabled. With this incidence of disability approximately two special school places are needed per 1,000 annual births. Knox examined the consequences of adopting either of two treatment policies: (*a*) early operation on children with no paralysis and no operations on other children—this would halve the number of disabled children at five years and reduce the number of special school places to about 1 per 1,000 annual births; or (*b*) early operation on all living affected children—this would approximately treble the number of disabled children and increase the requisite special school places to about 7 per 1,000 annual births. The number of places in special schools could of course be reduced if ordinary schools were adapted to accommodate those patients whose mentality was normal.[6]

Incidence of Meningomyelocele

Meningomyelocele, with or without hydrocephalus, is not one of the diseases notifiable under the Public Health Act and Regulations, but is notified on a voluntary basis to medical officers of health combined with compulsory notification of birth. The information so gathered is centrally reviewed by the Registrar General's Office*. They estimate that in 1967 this condition occurred in 1·47 live births in every 1,000 in England and Wales.[7] The calculations made in this paper have been based on this figure. The population of England and Wales in 1968 was 48,593,000, with a live birth rate per 1,000 population of 16·9.[8]

The total annual incidence of meningomyelocele, with or without hydrocephalus, for England and Wales in 1968 may therefore be estimated as 1,207 patients. A nationwide policy of primary closure would result in about 930 survivors for two years and 500 beyond this. If an average expectation of life of the latter group is assumed to be 40 years the total number would eventually be 20,000. For comparison Gehrig and Michaelis[9] reported that the annual incidence of traumatic paraplegia and tetraplegia in Switzerland is 15 per 1,000,000 population. In England and Wales this would give a total of 727 patients a year, provided the incidence was the same as in Switzerland. Thus the adequate treatment of meningomyeloceles is almost doubling

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*Registrar General's Office = Office of Population Censuses and Surveys.

the incidence of paraplegics annually. In the poliomyelitis epidemic of 1947-58 the numbers in England and Wales of permanently incapacitated patients accumulated, reaching a peak in 1957 of 7,700. The accumulated incidence for meningomyelocele in a similar period would be about 5,500 patients, and for traumatic paraplegia 7,997.

Medical Services Required

The Sheffield experience[1] shows that it is essential to establish a set area from which referrals can be accepted. After this area has been delineated all patients are accepted within the first 24 hours of life for closure of the defect. During this admission half of the patients will require, in addition, the insertion of a valve to decompress their hydrocephalus. The valve needs to be changed at least once every five years. An intravenous pyelogram is performed and a full orthopaedic assessment of muscle function in the legs is undertaken.

At about 4 weeks the child is discharged home. This may seem early but it helps parental acceptance and eases pressure on the nursing staff. The primary phase of the patient's care in hospital requires the services of an adequate neonatal care unit. After discharge the patients are reviewed in combined clinics staffed by representatives from all the disciplines involved in managing the child. The patients are reviewed at three months, six months, nine months, one year, and then every six months until they stop growing. Any discipline may, of course, need the child to be seen more often. It is advantageous that one person should keep the general state of the patient constantly under review. In Sheffield this is the paediatrician, who may be called on for general assessment before any operation.

In a paper such as this it is of no consequence to give the times at which some of the many operations should be performed. It is, however, important to point out that each child will require about one operation a year until the age of 5 and that half of them will also require valve revision at least every five years. When they stop growing 11% of spina bifida cystica patients are in wheel-chairs.[2] Lorber[10] felt this to be a very low estimate for meningomyeloceles and that 50-60% was nearer the truth. After completing formal education about 70-75% of patients can work in some capacity.[1] The remainder cannot work and require permanent care.

A service of this kind requires a neonatal care unit plus beds to support the inpatient requirement when further operations are needed. Such a unit depends on its local social services for rehabilitation and aftercare and on its local councils for education and housing for the patients. Outpatient and investigatory facilities will, of course, also be required.

Cost of Medical Services

Thirty per cent. of all children born with this malformation will die irrespective of treatment. Most, however, will not die immediately at birth and will therefore require a period of care. Thus nationally 1,207 patients will require nursing in a neonatal unit each year, and about 930 survivors will need treatment for two years and 500 subsequently. Centres undertaking this care may well not have such a neonatal unit, and the cost of establishing a unit with 15 cots is £64,000.[11] The initial stay lasts one month. Therefore one cot serves 12 patients. Hence 100 cots will be needed in England and Wales costing about £100 a week: a total annual running cost of about £500,000.

Every child requires at least one operation a year for the first five years of life. Thus 3,360 operations will be required each year. After the initial stay of one month, if the average stay in hospital is assumed to be two weeks, one bed will serve 25 children, and 138 beds will be required for England and Wales each year. The cost per inpatient case in London teaching hospitals in 1969 was £131·09. In provincial teaching hospitals in England and Wales this figure was £96·83.[12] Since most if not all of these children will be treated in a teaching hospital this approximate average cost will be £110 per bed per week. Thus each year the cost would be £800,000 for inpatient hospital treatment. This figure does not take into account the cost of treating those patients over the age of 5 years who need admission to hospital. Sharrard[1] estimated that in the Sheffield Region 70 beds are required to manage the service commitment to which 150 new patients are added each year. Thus from birth to about the age of 16 each child will require half a bed a year. This would bring the total annual cost of treating these patients to £1,430,000.

Social Services and Their Cost

These children will require education—some in special schools, and some, whose mentality is normal, at normal schools. The total figures for spina bifida cystica indicate that a high proportion of patients will attend normal schools, but for mengingomyelocele a greater proportion will have to attend special schools. In 1968-9 the current annual institutional expenditure for each normal pupil from public funds in England and Wales was £91 at a primary school. Pupils under 16 at a secondary school cost the Exchequer £171 each. The figure for a handicapped child in the same period was £423.[13] All these figures had increased by 50% since 1963. Suppose half (250 patients) attend each type of school. Children are at school for 11 years so 2,750 patients will be attending each type of school. Children leave primary school at 12 years of age, so the cost of each group will be £262,250 and £1,163,250 respectively. In contrast, if no initial treatment were to be undertaken only 6% of children with meningomyelocele would survive. If all these were educable, which is unlikely, the cost per year would be about £100,000. The cost of treating and educating these children who receive no initial surgical treatment, but who receive such treatment as is now ethically thought to be necessary, is about half that of educating those patients who are treated vigorously from birth.[1] Even then the cost of this last group to the Social Services will still be appreciable: about £700,000.

Centres are needed for the care of children after school at the end of the day and for adults. An estimate of the institutional cost for these children has been obtained from a children's home, whose management wish it to remain anonymous. The cost per child per week ranges from £26 to £37. If a quarter of these children live in children's homes the annual cost would therefore be about £370,000.

Overall Cost in the Future

From these calculations it appears that £$3\frac{3}{4}$ million a year is a rough estimate of the cost of treating, educating, and housing these children in England and Wales. This sum is a minimum: it does not include the cost of building such specialized departments as a neonatal unit, the cost of outpatient and investigatory facilities, ambulances, district nursing, and appliances, or the cost of building homes for dependant adults with the condition and thereafter supporting them.

All these estimates are almost certain to be exceeded in future. In the decade 1950-9 the cost of a hospital bed in England and Wales doubled.[14] There is no reason to believe that this increase will not be maintained over the next decade. The cost of school places is likely to rise in the same proportion, so that in 10 years' time to treat and educate these children may cost £$7\frac{1}{2}$ million a year. Against this figure the economic benefit is that represented by the earnings, necessarily limited, of the 70-75% of these children who may eventually work.

Legal Cost of Life

For comparison with these figures it is interesting to note that the maximum award generally accepted by the legal profession for accidental death of a person without dependants is £500. Should the deceased have dependants the awards vary greatly, each being assessed on the dead person's earnings when he was alive and his normal life expectancy. If a child with meningomyelocele were to be killed in an accident it is unlikely that his estate would receive an award exceeding £125. Awards for injury are much higher. A boy of 16½ rendered partially quadriplegic by an accident has been awarded damages totalling £62,500.[15] The awards to the victims of the thalidomide tragedy varied greatly, depending on the disabilities sustained, but the settlement figures ranged from £12,000 to £25,000. These figures may be compared with the average cost of saving the life of, and subsequently treating, a patient with meningomyelocele of about £7,250 (= £3,725,000 ÷ 500).

Conclusions

Early closure of a meningomyelocele leads to an increase in the number of patients who survive. The cost of maintaining a unit to care for these patients medically, to educate the survivors, and to house them is estimated, as is the future cost. These figures (which are approximate) apply only to England and Wales, and are £1,930,000, £1,425,500, and £370,000 respectively. The total cost might be expected to rise to at least £7½ million in 10 years. Though not all the possible patients will be referred to a new unit it can be seen that the cost incurred would be far beyond the scope of the funds available to the National Health Service if other and possibly more necessary commitments are to be honoured. If such units were to be established more funds would have to be allocated. Failing this, money from private sources would have to be sought. The Spastics Society's accounts[16] show that they raise about £2,000,000 a year. There is a suggestion that such a sum is becoming increasingly difficult to acquire, and as expenses rise the money is financing much less. Such a cost brings into question once again whether early closure is warranted. Ethically many people believe that it is, and therefore the search for funds becomes essential. It would, however, seem to be undesirable to promote a nationwide policy of early closure of meningomyelocele until the funds are available, since to produce survivors who are then uncared for would be worse than to fail to produce survivors at all.

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Clinical Problems

Nephrotic Syndrome in the Elderly

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Summary

Though the nephrotic syndrome is generally believed to be uncommon in the elderly, patients aged 60 years or more accounted for 25 out of 100 consecutive adult cases. Six (24%) of these had the minimal change lesion, compared with 16% of the younger adults. The incidence of membranous glomerulonephritis was similar in the two age groups, but proliferative glomerulonephritis was more common in the younger (29%) than in the older group (16%). Amyloidosis did not have a higher incidence in the higher age group. Five of the elderly patients with minimal change lesion were treated with prednisone—in four a complete remission from the nephrotic syndrome followed, while the fifth patient's course is unknown.

These results suggest that, when the patient's other circumstances allow, the nephrotic syndrome in an elderly patient should be investigated and managed as in younger age groups.

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