Middle Articles

Spina Bifida Cystica and Family Stress

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The number of infants born alive with spina bifida cystica in England and Wales is about 1,500 a year. Until a few years ago most of them died within a month or two of birth, but now, through surgical advances, nearly one-half survive for a year and nearly one-third for five years (Laurence, 1964). There will be an increasing need for medical and social services to deal with the handicaps commonly associated with this malformation (paralysis and deformity of the legs, incontinence of urine, etc.), and an important part of such services should be the provision of aid and advice to parents, on whom the strains, both physical and psychological, of caring for the children are often very severe.

We set out to study the nature of the stresses imposed on a family by the birth of an infant with a major malformation of the central nervous system—that is, spina bifida cystica, anencephaly, congenital hydrocephalus without spina bifida cystica—and to see how far the existing medical and social services were adequate. Below we report some of our preliminary results and conclusions after two years' work.

Method and Case Material

The study developed from a socio-genetic survey of malformations of the central nervous system in South Wales (Laurence and David, 1964, 1966; Laurence, 1966; Carter, Laurence, and David, 1966), and the present report is based on our first 120 cases, notified to us between March 1964 and November 1965 and studied to the end of 1965. The primary malformations in these cases were: spina bifida cystica, 97; anencephaly, 20; congenital hydrocephalus, 3. In each case the mother was interviewed as soon as possible after notification (usually within three days of the birth). One month after the birth both parents were interviewed at home, and the mother was visited again at six months. Thereafter, where the child survived, the mother was visited at six-monthly intervals. No mother refused the initial interview, but eight cases were later closed through refusal, and in three cases the family moved from the district.

When a mother bears an obviously malformed child severe emotional distress is bound to be in store for her and her husband. First, the parents must be told of the abnormality and some attempt made, then or later, to explain its nature,

cause, and consequences. If surgical treatment is required, as in most cases of spina bifida cystica, the mother will have to suffer her baby being taken from her and may not see it more than fleetingly for another fortnight or so. Back at home she will have a delicate child with an operation wound to care for, and it will often be obvious to relatives, if not to the mother, that the child is abnormal and its life precarious. At some time during the first few days the parents are likely to be told of the risk of meningitis, of the need for further treatment (of hydrocephalus, orthopaedic deformity, or incontinence), and of the possibility or probability that the child's legs will be paralysed and that it may never walk. Frequent visits to hospital may be necessary. Another source of worry will be the possibility of further pregnancy. If the parents are anxious to avoid or postpone another pregnancy they may find themselves uncertain about contraceptive measures; if they want more children they will be concerned about the risk of another child being affected in the same way. We now consider the attitudes of the parents to some of these problems.

Telling the Parents

At the first interview parents were asked who told them of the baby's malformation, what they were told, and what was their first reaction. Their replies appeared to indicate that fathers were more distressed than mothers by the news, but it became evident that mothers, at the time of the first interview (usually only a few days after the birth), had not really taken in the situation.

Case 118.—Spina bifida cystica. Mother aged 32; second child, hospital delivery. At the interview, three days later, the mother said she was told about the baby's condition by a nurse a few minutes after delivery. She was still partly under the anaesthetic, but thought they said there was something wrong and that they had to operate. She couldn't take it in really, but was very upset. She had so many different stories from the nurses and sister that it took her a long time to realize that they were talking about all babies of that kind and not about her baby in particular.

At the second (one-month) interview parents were asked whether they thought a mother should be told about the baby's condition at once or later. The great majority of mothers (90 out of 99) said she should be told at once, but one-third of the fathers (33 out of 96) thought she should be told later, either by the father or at the same time as the father. Parents who thought a mother should be told later generally based this on the opinion that she was not really in a fit state to take things in.

Case 33.—Spina bifida cystica. Mother aged 25; fourth child. She said the father should be told first so that he can tell the mother, or else the parents should be told together. When she was

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told about her baby (a few minutes after delivery): "It shook me, but I didn't take it in until I saw my husband and he tried to explain."

Case 118.—Spina bifida cystica. Mother aged 32; second child. Mother couldn't say when you should tell. If you didn't tell at once you would give false hope. On the other hand, if it was said too early, as in her case, the mother couldn't understand what was being said.

Parents were also asked at the one-month interview how they felt about the way the news had been broken to them; 80% thought this had been done in as kindly a way as possible, 8% had some reservations, and 12% felt it had not been well done. Breaking the news to parents in such circumstances is, of course, a very difficult and delicate matter. We record here some of the parents' remarks and offer comment in the next section.

Case 7.—Anencephaly. Father 28, mother 27; second child. Father was told over the phone that the baby was stillborn and that the child's head was not properly formed. He thought this was not well done. It hurt him being told over the phone; somehow it seemed he was given the news secondhand. Also two other people, a relative and a neighbour, were told about his wife having a malformed stillborn baby over the phone by the hospital staff. The hospital staff were courteous, but he felt he was not being treated properly, not having seen any doctor or had any explanation given him.

Case 38.—Spina bifida cystica (mild). Mother 16; first child, hospital delivery. Two days after the birth the mother noticed a mark on the baby's back which she hadn't seen before. Later the doctor told her about it. He said the baby might be paralysed and might get meningitis. "It was awful brutal." To her he seemed to be shouting, though she agreed she may not have been in a fit state. At the one-year interview she talked of the "terrible" time she went through when the baby was born; if only somebody had told her husband first, then he could have told her instead of being given such a shock and then having to explain to her husband about the baby.

Reactions of Relatives and Neighbours

We had expected that parents might often be distressed by uninformed or thoughtless remarks from relatives and neighbours, but in fact this was rare. The great majority of parents said they did not mind people knowing of the baby's condition and that friends and relatives had all been sympathetic and helpful.

Case 83.—Spina bifida cystica. Father 27; first child. Father said that at the beginning he had tried to avoid people, but as soon as neighbours in his street knew they were so terribly kind to him in every way. He could have gone into any house and everybody would be extraordinarily kind.

Case 8.—Spina bifida cystica. Mother 17; first child. Mother said no one had made unkind remarks, but her mother-in-law likes to show other people the baby's back. Mother resents this very much, but is too shy to say anything.

One problem that parents have to face is that of relatives' remarks, and their own thoughts, on whether it would be better if the baby died. Almost every mother wanted her baby to live, especially when she had had it home and looked after it. Our impression was that mothers were anxious and unsettled while the baby was in hospital (for surgical treatment) and happier when they had the baby home, even when it was obviously in a poor state. Many of the babies were wizened and feeble, and, with developing hydrocephalus, often looked unnatural or even grotesque to other people, but the mothers did not seem to see their babies in that way and continued to love them and want them.

Case 78.—Spina bifida cystica with severe hydrocephalus. Father 24, mother 21; first child. Father said that when he saw the baby (with hydrocephalus) he wished they wouldn't continue to keep the child alive. If the future was as bad as they had painted it he would much rather the baby didn't live. But he was surprised

how concerned everybody was and how good and kind they were. Mother said that all she wanted was to see and have her baby.

Case 8.—Spina bifida cystica. Parents aged 17; first child. Mother had been told the baby was never likely to walk. At the first interview she said she would much rather the baby died, as if the child were a cripple she wouldn't know how to look after her. Father intervened with, "She doesn't love the baby yet as she should; she hasn't handled her much, but she'll come to it." At the six-month interview the mother was reported to be "terribly fond of her baby."

Effect on Marital Harmony

The emotional and physical strain of having and caring for a malformed child is likely to influence marital harmony in a way that reflects the depth of affection between the parents and the maturity of their personalities. In a great majority of cases, and whether the child survived or not, parents said that the event had brought them closer together. In only one case was the baby neglected by both parents and had to be taken into the care of the children's department; here the mother, aged 35, was Italian and spoke English poorly, and the father had been invalided from the Services with neurosis. There were three cases in which the mother's preoccupation with the child or the father's inability to accept it led to quarrels and separation.

Case 32.—Spina bifida cystica. Father 24, mother 21. Fourth child (second child stillborn). Mother said her husband was very nervous of the baby, and it seemed as if he had more or less disowned responsibility for the child. At the one-year interview, the mother said she and her husband had had "murder." He blamed her for the malformation and she blamed him. She made the percipient remark, "Sometimes you have to hurt people because you are so upset and worried."

Enough Help and Advice?

Parents were asked at one month, and mothers again at six months, if they felt they had been given enough help and advice by the hospital, their general practitioner, and their health visitor. One-third of the fathers and more than a half of the mothers felt they could have been given more help or advice by one or more of these three sources. These parents appeared to feel, not so much that they should have been given more instruction, but that the doctor or nurse didn't seem to appreciate what worries and anxieties the parents had to bear and didn't say the simple words of comfort which would have indicated their understanding and sympathy.

Case 2.—Spina bifida cystica with hydrocephalus. Mother 27; first child. Mother felt parents should be told more. "The hospitals don't tell you much. They would not sit down and explain to you."

Case 24.—Spina bifida cystica. Mother aged 35; third child (second child also spina bifida, lived one day). Mother said there should be less confusion at the hospital and more definite answers. There should be explanations why things were being done. The doctor sent her to the surgeon, but he didn't tell her why. The baby had a kidney radiographed but no one told her if it was all right. She felt they were being treated like a lot of animals, herded here and there.

Visits to Hospital

At the one-year interview mothers were asked about their visits to hospital. The malformed child, if it survived, nearly always needed observation or treatment necessitating either in-patient care, during which the mother would visit frequently, or outpatient attendance, when the mother would have to take the child to and from hospital. Of 23 mothers whose children were living at one year only four received partial assistance from the ambulance service for journeys to hospital; six went

by car, the rest by bus or train. Fourteen mothers visited hospital at least weekly during the first three months. The cost of the journeys varied from "negligible," where the mother went in the family car, to 20 shillings, where the mother went by train with a relative. The average cost of all journeys was about 7s. 6d. a time, always paid by the family. The time taken by the journey each way varied from 10 minutes to two hours, averaging about an hour, and the average time spent at the hospital was about an hour and a half. Thus a visit to hospital tended to occupy a large part of a day, and the cost, over the months, was such that some husbands said they worked overtime to earn the extra money.

Risk for Further Children

Mothers were asked whether their child's condition had affected their attitude to having further children, whether they thought there was a risk that it might happen again, and whether anyone had told them about such a risk. At the one-month interview a third of the mothers said they would definitely not want any more children, and one-fifth said they would be cautious. These proportions were somewhat less at the six-month interview. We did not ask specifically about contraception, but it became evident that few mothers had much knowledge of this. Among 35 cases under observation for a year four mothers were known to be pregnant again, two unwillingly.

One-quarter of the mothers said they thought there was no risk that the same trouble would happen to them again, and one-quarter did not know whether there was a risk or not. Of the mothers who thought there was some risk the majority (two-thirds) either did not know how great it might be or assessed it as "small." In reply to the question whether anyone had talked to them about a risk, 60% of the mothers said "No" at the one-month interview, and the proportion was the same at six months. Of those told about a risk by their doctors the general impression received was that it was small. Parents said they were told: "it could not happen again," "there was a one-in-a-million chance," "one chance in 50,000," "extremely unlikely," "a slight chance."

Case 5.—Spina bifida cystica. Mother aged 17; first child (died after four days). "My doctor said it was a bit of bad luck and there was no likelihood of it happening again." The mother wants another baby and isn't trying to avoid pregnancy, but says she would be very afraid in case it went wrong again.

Case 23.—Spina bifida cystica. Mother aged 20; first child (alive at one year). Father says his general practitioner told him there was one chance in a thousand it would happen again. Mother says the doctor told her it would be impossible for her to have another baby with the same trouble, but she met someone in hospital who had had two spina bifida babies.

Discussion

It will of course be appreciated that our information was obtained largely from parents who, under the stress of emotion, may have misapprehended some of what was told them by doctors and nurses. Moreover, where parents express dissatisfaction with the way their case was managed this may in part be due to the psychological process by which feelings of anxiety or guilt are converted into expressions of hostility and blame. The question of when parents should be told of an abnormal birth and how much should be said of its nature and causes must be decided by the doctor or nurse from their experience and from an assessment of the particular circumstances. The great majority of parents (four-fifths) felt the news was broken in as kindly a way as possible. Where this was not so it often seemed as if the anxiety aroused in the doctor or nurse at having to impart distressing news had itself been a cause of clumsiness or oversight. Training as well as experience could help guard against this.

Our findings that many mothers seemed unable to take in what was told them soon after delivery suggests that it may often be wiser to break the news only in the most general terms at first and leave more precise explanations till later, perhaps when the husband can also be present. In this and other ways our findings agree with the experience of Franklin (1963).

Once at home with their babies most of the mothers had the sympathy and help of relatives and neighbours. Yet at the same time mothers were apt to feel on their own, carrying responsibilities of which those around them had no experience. For this reason they especially looked for advice from their health visitor and general practitioner, and easily became discouraged if these persons did not know of, or showed no special concern for, their child's disabilities. Mothers were often eager for an opportunity to talk to others who had faced the same problems, and several asked if we could put them in touch with such mothers. This led us to think a parents' association would be appreciated, and the interest shown by parents in the South Wales Association for Spina Bifida and Hydrocephalus, inaugurated October 1965, seemed to confirm this.

Any consideration of the welfare of families with a severely handicapped child must take into account the presence of other stresses in the family life. The emotional strain of accepting the abnormality, the anxieties of nursing a delicate or fretful child, and the fatigue of frequent hospital visits are common to most of such families. But ill-health in the parents, the needs of other small children in the family, a father's unemployment, unsatisfactory housing conditions, or living with in-laws may all increase the natural burden, while neurotic traits or immaturity of personality in one or both parents may increase it past endurance. Yet the number of such families in any particular community is not, after all, very great, and a health visitor or welfare worker in the course of her routine duties is unlikely to gain much experience of their particular problems. Moreover, as the problems are often manifold and complex, assistance may be sought from several different departments of the local authority or voluntary organizations. This carries the risk of duplication and conflicting advice; as Kendall and Calman (1964) have expressed it, there are apt to be "too many fingers in the pie, but no cook." It has seemed to us that most benefit would result if one person, a worker with special experience in the field, were to act as co-ordinator for the provision of social and medical help to each such family over an appropriate area. She would be the person who has close knowledge of the problems in each family and to whom the parents would always turn for advice. We think, too, there is a strong case for special hospital centres to provide major surgical and orthopaedic treatment for all cases within an appropriate region.

Previous studies of family problems in relation to congenital disorders have stressed the need for adequate genetic counselling (Tizard and Grad, 1961; Kendall and Calman, 1964). Our own findings confirm this need. When a mother has had one child with spina bifida cystica or anencephaly, the risk of recurrence is undoubtedly a serious one. The recurrence rate was found to be one in 33 in Birmingham (Record and McKeown, 1950), one in 17 in Rhode Island, U.S.A. (MacMahon *et al.*, 1953), one in 18 in Southampton (Williamson, 1965), and one in 16 in Sheffield (Lorber, 1965). In South Wales, where the present study is being made, the recurrence rate after a spina bifida birth is one in 15, and after an anencephalic birth, one in 25 (Carter and Laurence, 1966).

Although a doctor, in advising parents, will need to consider how far a realistic statement of the recurrence risk may lead to undue anxiety, yet in general, and particularly where parents ask for information, it will be proper for him to quote the best evidence from specialist studies, or else to refer the parents to a clinic for genetic counselling. If, on the basis of such information, the parents decide they want to avoid further pregnancies, at least for the time being, then there are good reasons why contraceptive advice and means should be medi-

cally available to them. Adequate contraception, by relieving a mother of her fear of another pregnancy with its attendant risk, is an important factor in the maintenance of marital harmony. It will also permit the parents to become accustomed to the present strains and the future problems of bringing up a handicapped child and so to take a measured decision on having further children.

Summary

This paper describes a study of the stress imposed on families by the birth of a child with a major malformation of the central nervous system (spina bifida cystica, anencephaly, congenital hydrocephalus without spina bifida). One hundred and twenty families have been studied, the parents being interviewed by a social worker soon after the birth and again at one month, six months, and a year. Some findings and conclusions from the first two years' work are reported.

The series contains 97 cases of spina bifida cystica. Of 45 families under observation for a year 22 of the children were known to be surviving.

The problems studied were those of telling the parents about the child's malformation, the reactions of the parents and relatives to the event, its effect on marital harmony, the adequacy of medical advice, the numbers of hospital visits, and information on the risk of recurrence in further children.

The findings suggest (a) the need for care, skill, and sympathy in telling both parents about the malformation, (b) the advantages of a parents' association, (c) the need for a worker with

specialized experience to co-ordinate the provision of social and medical help to the families, (d) special centres to deal with the medical and surgical problems of all the affected infants in an area, and (e) the need for adequate genetic counselling and advice on contraception.

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HOSPITAL TOPICS

Hospital Building Programme

The following statement was issued by the Ministry of Health last week

Recent comments on the amount of capital expenditure for hospitals have criticized the present position as being, in proportion, below the England and Wales prewar level and well below the current level in other countries. The contention that the situation is both worse under the National Health Service than formerly, and deteriorating, is not, however, borne out by the facts. While no one would dispute that a considerable distance has yet to be travelled before all the needs of a fully efficient Health Service can be met, progress has been and is being made in that direction. The rate of progress may be slower than we would wish, but it is conditioned by national resources of money and manpower for which there are many claimants. The health share of resources is nevertheless increasing.

Comparison with the Pre-1939 Position

Precise comparison is not possible because hospital building in the 1930s did not proceed under any co-ordinated plan and was financed from many different sources. The available information is incomplete, but it

has been estimated that annual expenditure on hospital building in England and Wales just before the second world war was of the order of £10m., or at present prices about £46m. This latter figure compares with a current level of spending of £73m. which is expected to rise to at least £100m. by 1970. Present expenditure is therefore nearly 60% above the 1938-9 level and the rate will soon be double that of prewar.

In 1938-9 hospital capital expenditure was 0.19% of the gross national product, in 1965-6 the proportion had risen to 0.23%, and on the basis of the National Plan it would rise still further to 0.31% in 1970.

Comparison with other Countries

An unpublished W.H.O. document provides the only available data for purposes of comparison. According to this source the total capital expenditure on health services (the major part of which will have been on hospitals) expressed as a percentage of the gross domestic capital formation was: Canada 2.31 in 1961, Sweden 2.12 in 1962, United States 1.76 in 1961-2, United King-

dom 1.18 in 1961-2, Australia 1.16 in 1960-1, and Czechoslovakia 1.10 in 1961. Although no later information is available for the other countries named above, U.K. capital expenditure on health services has increased very sharply in real terms since 1961-2. In any event the W.H.O. figures lend no support to the allegation that in proportion Sweden is spending ten and a half times as much as Britain, and Czechoslovakia eight and a half times as much.

Achievement in First 17 Years of National Health Service

In the 17 years from July 1948 to September 1965 the total amount spent on completed schemes for hospital building was nearly £337m. About half of this sum related to completions in the last four years of the period. In the earlier years the capital available was small—an annual average of about £9m. between 1949-50 and 1954-5. The mid-1950s saw the beginning of growth and the amounts spent on hospital building increased steadily from £10m. in 1954-5 to nearly £24m. in 1960-1, rising even more