arm-swelling and raised venous pressure, *x*-ray treatment, axillary metastases, arterial blood supply, or use of the arm. Wound infection and axillary scarring were the main factors in their opinion.

It is difficult to draw conclusions from this contradictory literature. But there seems to be no justification for denying that venous obstruction is a possible cause. This is not necessarily a direct result of axillary scarring, since Wyburn-Mason (1953) demonstrated the roles of costo-clavicular compression in four patients whose arm-swelling could be relieved by a sling support or excision of part of the first rib.

Post-operative and/or post-radiation scarring in the axilla is probably the most common factor, however, and the best conclusion from the present evidence is that of Aird (1955) that arm-swelling is "due either to the blockage of axillary lymphatics by malignant tissue, or to removal of the axillary lymphatic bottleneck in a radical amputation of the breast or to the fibrosis that follows high-dosage irradiation of the axilla."

Conclusion

This study has confirmed the clinical impression that arm-swelling often occurs after treatment for breast cancer. A morbidity rate of 36% can be expected when the method of treatment is radical mastectomy and post-operative radiotherapy, including the axilla. This is not to suggest replacing radical by local mastectomy, nor dispensing with *x*-ray treatment to the axilla. The survival rate is more important than the morbidity rate. But it might be rewarding to make a further study, as outlined above, to try to find out if a less radical mastectomy and a less severe axillary radiation reaction would be likely to reduce the morbidity due to arm-swelling without reducing the cancer survival rate.

Summary

Arm measurements were made of 305 unselected patients after routine treatment for breast cancer—a significant degree of arm-swelling was found in 36%. This morbidity rate is discussed.

Evidence was collected of the patient's subjective awareness of arm-swelling, and 55% complained of some inconvenience from this symptom. Significant swelling was found in 50% of patients whose axillary lymph nodes were involved with cancer, and in 28% of those which were not.

It was found in 46% of patients who had received *x*-ray treatment to the axilla and 14% who had not.

In 41% it was found after radical mastectomy, in 12% after local mastectomy.

There was no significant difference between left or right arm involvement.

The aetiology of arm-swelling is discussed.

I am grateful to the surgeons of the United Oxford Hospitals whose patients form the basis of this study. In particular, I thank Mr. A. Elliot-Smith, who has read this manuscript and made several helpful criticisms.

This study was carried out in the radiotherapy department, and I thank the Director, Dr. Frank Ellis, for his advice in the preparation of this manuscript, and the other members of the staff for their help in the collection of the data.

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"PREMATURE" CHILDREN AT PRIMARY SCHOOLS*

BY

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In an earlier paper (Douglas, 1956a) I described the mental test performances of 8-year-old children of low birth-weight. These "premature" children made consistently poor scores in both mental-ability and school-achievement tests when compared with a group of carefully matched controls. Their scores, unexpectedly, showed no clear relationship with either birth weight or length of gestation. Relatively high scores were made by those premature children born of mothers with a history of toxaemia or other abnormality during gestation, whereas the absence of such a history was associated with low scores. No adequate explanation could be given for these findings.

Further tests were given to the same groups of premature and control children when they were 11 years old, and the results of their 11+ secondary selection examinations are also known. In addition, their teachers have reported on their powers of concentration, discipline, and attitude to work. This new information is described in the present paper.

Background to this Study

A unique opportunity to make a controlled study of a representative group of premature children was provided by the Maternity Survey of 1946 (R.C.O.G. Survey, 1948). All children born during the first week of March of that year were enrolled in the inquiry, and among them were 706[1] legitimate single-born babies

*This inquiry is being made by a Joint Committee of the Institute of Child Health (University of London), the Society of Medical Officers of Health, and the Population Investigation Committee. The chairman of the Committee is Professor James Young, the vice-chairman Professor A. Moncrieff, and the secretary Professor D. V. Glass. The Nuffield Foundation has taken a major part in financing this work and grants have also been made by the Board of Governors of the Hospital for Sick Children, Great Ormond Street (through the Institute of Child Health), by the Ford Foundation, by the Population Council (Inc.), and by the Medical Research Council. Fourteen regional hospital boards have also made grants to the inquiry from their free moneys.

[1] The number given in earlier papers was 707, but one child has since been excluded.

weighing 5½ lb. (2.5 kg.) or less at birth, and therefore, by international definition, "premature." Information was lacking on the home conditions of 31 of these premature children, but we have been able to match each of the remaining 675 with a child (the control) weighing more than 5½ lb. at birth who was selected from the remaining legitimate singletons (some 12,000 in number) born during the survey week. The selection was made on the basis of sex, ordinal position in the family, mother's age, social group, and degree of crowding in the home; and wherever possible the premature child was matched with a control living in the area of the same local authority.

Losses.—During the first 11 years of this study *unavoidable* losses through death and emigration have been heavy. In addition, as will be seen in Table I, a number of parents refused to co-operate, and some children were unable to take the tests owing to illness or mental backwardness. The greatest source of *avoidable* loss in the 11-year tests was late testing. The test booklets did not reach some schools until just before the holidays and were not completed until the beginning of the next term. This error of administration does not appear to have introduced a bias into the 11-year test scores, since the children excluded had 8-year test scores close to the average of the group from which they came.[2] As in previous papers, the absence of information on one member of a pair excluded the other member also from the analysis.

TABLE I.—*Losses from the Matched Sample (Birth to 11 Years)**

		8-Year Tests	11-Year Tests
Unavoidable losses	Death	178	179
	Emigration	36	39
Avoidable losses	Refusals	19	25
	Late or untraced	18	50
Inability to test	E.S.N. or M.D.	8	17
	Ill or physically handicapped	8	10
Completed all tests		408	355
	Total in sample	675	675

* These figures differ somewhat from those given in 1956. Owing to a clerical error some losses were attributed to emigration which should have been attributed to refusals or failure to trace. In addition, one child who completed only three out of four tests has been excluded from the analysis in this paper, whereas two other children whose test scores were not available in 1956 have been included.

Description of the Data Used

The tests used were designed for this inquiry by the National Foundation for Educational Research in England and Wales. They consisted of four tests given when the children were 8 years old and another four when they were 11 years 3 months. The tests given at 8 years were a picture intelligence test and reading, vocabulary, and sentence-completion tests. At 11 years the tests given were a mixed verbal and non-verbal intelligence test, and tests of reading, vocabulary, and arithmetic. The identical reading and vocabulary tests were used at each age. Both batteries of tests were given to the children by the primary-school teachers or, in a few instances, by educational psychologists.

The crude scores achieved by children in these tests have been converted into T scores based on all children at the particular age tested with a mean of 50, and a standard deviation of 10. This conversion was not used in the 1956 paper, and, as is shown below, has slightly altered the findings.

[2] The average test score at 8 years of premature children who were not tested at 11 years was 48.2 and the 8-year score of those tested in both years 48.0.

In addition to the two batteries of tests given by the teachers, the results of the 11+ secondary selection examinations were supplied by local education authorities. Children in Scotland were excluded from this part of the study because of the different method of secondary selection practised there, and a few children living in areas of England and Wales where 11+ examinations were not held were also excluded. Children going to private schools (10 premature and 19 controls) could not, of course, contribute to this part of the study, though they were included in the mental test studies, and we were left with 309 pairs, both members of which took the selection examination for local authority secondary schools in England and Wales.

Another view of the educational progress of these children was given by their teachers' comments on their attitude to work, powers of concentration, and discipline. These were made in response to questions put to the teachers when the children were 10 years old.

Lastly, special records of school absences were kept by the primary-school teachers.

Results

The main purpose of this paper is to describe the progress of premature children attending normal primary schools. In addition, six premature children and two controls were described as educationally subnormal or mentally defective at 8 years, and 11 premature children and six controls at 11 years.[3] The inclusion of these severely handicapped children, if we had been able to test them, would not have altered the picture given below, since they are few in number and are not confined to the premature group alone.

The test scores of the premature and control children are shown in Table II.

TABLE II.—*Test Scores in Tests of Mental Ability and School Achievement at 8 and 11 Years*

	Controls	Prematures	Difference
Tests at 8 years (408 pairs):			
Sentence completion	50·9	47·9	−3·0*
Reading	51·0	48·0	−3·0*
Vocabulary	50·0	48·3	−1·7*
Picture intelligence	50·5	47·7	−2·8*
Tests at 11 years (355 pairs):			
Reading	50·1	47·1	−3·0*
Vocabulary	50·3	47·3	−3·0*
Arithmetic	50·4	46·6	−3·8*
Mental ability	51·1	46·9	−4·2*

* Each of these differences is statistically highly significant when tested by Student's t-test.

The scores of the controls were consistently a little above the level to be expected—that is, 50—and the scores of the premature children consistently below it. The scores for the latter were lower at 11 years than at 8, whereas the control children made similar scores at each age. Thus, the premature children appear to become more handicapped as their age increases. They were similarly handicapped in all tests, and the suggestion made in 1956 that they were particularly backward in reading cannot be sustained now that test scores are used.

The premature children also did badly in the 11+ selection examinations. Only 9.7% gained a grammar-school place, as compared with 22.0% of their controls.[4]

[3] The figure of 11 premature children and six controls, who were reported as educationally subnormal or defective at 11 years, is a minimal one, since the comments of teachers show that some children, both premature and control, attending primary schools should in their opinion have been allocated to special schools.

[4] A highly significant difference.

TABLE III.—*Success in the 11+ Secondary Selection Examination Related to Mental Test Score*

11-Year* Test Score	Premature Children		Controls	
	G. S. Allocation %	Total in L. E. A. Schools	G. S. Allocation %	Total in L. E. A. Schools
< 40	Nil	63	Nil	29
40–	Nil	46	Nil	51
45–	Nil	66	4·0	50
50–	1·9	54	12·5	72
55–	41·2	34	46·0	50
60+	70·6	17	73·2	41
Unknown	10·3	29	25·0	16
All children	9·7	309	22·0	309

* Average score on the four tests given at 11 years.

Within similar ranges of mental-test scores, however, the premature children were not at any substantial disadvantage (see Table III). The apparently better 11+ performance of the controls with test scores between 45 and 54 is based on only a few grammar-school places and is not statistically significant.

The results of the 11+ examination sometimes depend on the teachers' reports as well as on examinations and tests. It is therefore relevant to note that the teachers themselves made more adverse comments on the premature children than on their controls when asked to assess their powers of concentration, attitude to work, and discipline. Of the former 31% were the subject of one or more adverse comments as compared with 20% of the latter.[5] As with 11+ examination successes, the teachers' comments were closely related to the tested mental ability of the children (see Table IV).

TABLE IV.—*Teachers' Comments* Related to Mental Test Score*

11-Year Test Score	Premature Children		Controls	
	No Adverse Comment by Teacher %	No. of Children	No Adverse Comment by Teacher %	No. of Children
< 40	36·7	79	34·4	32
40–	65·0	60	75·4	61
45–	71·3	87	74·3	70
50–	81·3	64	86·0	86
55–	89·6	48	90·2	61
60+	100·0	22	98·2	55
Unknown	72·4	29	75·0	24
All children	68·9	389	79·7	389

* Comments on attitude to work, power of concentration, and discipline.

To summarize, all four sources of information—mental tests at 8 and 11 years, the results of the 11+ selection examination, and the teachers' comments—show premature children as being considerably handicapped. The reason for this is discussed below.

Explanation of the Poor School Achievement of Premature Children

One possible explanation—that the premature children were more often ill and so missed school—may be immediately dismissed on the evidence of the school absence records. Between the age of 6 and 10 years they were away from school for an average of three weeks and one day per year whereas their controls were absent for exactly three weeks.

In an earlier paper (Douglas, 1956b) I was able to show that an apparent retardation in walking and talking among premature children was satisfactorily explained when their age was calculated from conception rather than from date of birth. But their poor performance in mental tests cannot be similarly explained, as will be seen from the following figures: children of birth weights of 5½ lb. (2.5 kg.) or less who were born within a week of term had an average 11-year test score of 46.9; those born one to four weeks early a score of 46.6; and those born *more* than four weeks early a score of 47.4.

It is unnecessary to discuss the relation between the level of test scores and the causes of premature birth, because the apparently poor performance of premature children in these tests is explained below in terms of the social and educational background of their families. As this survey progressed it became increasingly clear that, in spite of the careful matching of the premature and control pairs at the beginning of the survey for fathers' occupations, there were, in fact, marked social differences between them which have grown larger with time. The fathers of the premature children were more likely to move during the survey period to less favourable occupations than the fathers of the controls, and were less likely to improve their occupational status or move into self-employment. Moreover, periods of unemployment were more than twice as frequent among them —6.5% were unemployed on one or more occasions as compared with 3.2% of the fathers of the controls.[6]

As previously noted, even when drawing controls from a parent sample of more than 12,000 children, matches were not always satisfactory. But this does not explain either the difference in occupational history or the difference in test scores. The latter were just as great in the apparently "perfectly" matched pairs (283 in number) as in those less well matched. I therefore made a more detailed study of the social and educational background of the parents of these children, their standard of child care, and their interest in their children's educational progress.[7] The results are shown in Table V.

TABLE V.—*Comparison of the Social and Educational Background of the Parents and Standard of Maternal Care and Interest in School Progress*

	Families of		Difference
	Premature Children	Controls	
Social and Educational Background:			
% of grandfathers in non-manual work: Maternal	26·7	29·4	−2·7%
% of grandfathers in non-manual work: Paternal	27·6	36·9	−9·3%
% leaving school after the age of 15: Mothers	15·1	18·9	−3·8%
% leaving school after the age of 15: Fathers	14·7	17·3	−2·6%
% taking night classes or other courses after leaving school: Mothers	23·2	24·6	−1·4%
% taking night classes or other courses after leaving school: Fathers	17·6	24·7	−7·1%
Assessments of Maternal Care and Interest in School Progress:			
% rated favourably by health visitor in 1950 for: Management of child	33·7	45·1	−11·4%
% rated favourably by health visitor in 1950 for: Cleanliness of child and home	41·8	53·9	−12·1%
% rated favourably by teachers for interest in the child's progress at school	29·2	41·8	−12·6%

The social and educational background of both the mothers and the fathers of premature children was consistently less favourable than that of the controls. More of them came from manual workers' families, fewer stayed at school after the age of 15 or attended

[5] Another highly significant difference.

[6] A highly significant difference.

[7] Two assessments of maternal care were made by health visitors when these children were 4 years old. "Management" refers to sympathy and understanding of child; "cleanliness" to the care of clothes and shoes and to the cleanliness of the child and home.

night classes or other courses after leaving school. These differences were more pronounced for the fathers than for the mothers, and there is no suggestion here that the mothers of premature children have poorer social origins than their husbands.

According to the health visitors, the mothers of premature children showed poorer standards of both care and management, and according to the teachers they also showed less interest in their children's school progress. These three assessments of care and interest discriminated more sharply between the parents of premature and control children than did either the parents' social origins or standards of education. The relatively low proportion of premature children attending fee-paying schools (10% as compared with 19% of their controls) is a further reflection on both the economic circumstances of the families and the interest shown in education.

Preliminary tables showed that the test scores of premature children were particularly low when they came from a family that was less favourably assessed for social origins, education, or parental attitude than their controls, whereas when they came from a family more favourably assessed they scored as well as or better than their controls. For example, when premature children had parents whose educational interest was greater than the parents of their controls they scored 6% higher in the 11-year mental tests. When the interest of both sets of parents was the same they scored 5% less, and when the parents were less interested than the parents of the controls they scored 16% less.

In order to show more clearly the extent to which differences in parental background and attitude contributed to the poor achievement of the premature children, each pair was marked according to whether the premature child was superior to (+1), the same as (O), or inferior to (−1) the control on each of the following items: (*a*) social and educational background of both parents, (*b*) maternal care and management, and (*c*) interest in school progress. When the premature child was superior in all three a mark of +3 was recorded and when inferior in all three a mark of −3. A mark of O was given to those pairs where the premature child was assessed less favourably than the control for one item and more favourably for another as well as to those pairs where assessments were the same for all items.

These environment and interest marks show clearly the extent to which the premature children were drawn from less favourable family backgrounds than their controls—48% were rated unfavourably in these respects, whereas only 27% were rated favourably. The distribution of marks is given in Table VI.

Table VII shows the test scores and 11+ selection examination results after grouping the pairs in this way.

The premature children were consistently better than their controls when they were favourably assessed and worse than their controls when they were unfavourably assessed. But they were still at a slight disadvantage in both tests and in the 11+ examinations when they were assessed similarly to their controls—that is, had O marks.

Table VIII summarizes the progressive reduction in the handicaps of the premature children as their parents' social origins and educational aspirations get nearer to those of the parents of their controls.

TABLE VI.—*Distribution of Marks for Social Background, Maternal Care, and Interest in School Progress*

	+3	+2	+1	0	−1	−2	−3	No. of Pairs Full Information
Distribution of pairs (%)	4·9	8·0	13·6	25·4	21·9	17·9	8·3	374
	26·5% Premature children more favourably assessed.				48·1% Premature children less favourably assessed			

TABLE VIII.—*Test Scores (Average of Four Tests at 11 Years)*

	Premature Children	Controls	Difference	No. of Children
All pairs	47·0	50·5	−3·5*	354
Pairs with same interest in school progress	48·2	50·9	−2·7*	136
Pairs with similar assessment of social and educational origin, care, and interest (i.e., O marks)	47·6	49·3	−1·7	79

* A highly significant difference.

The same picture is found for the 8-year tests and the 11+ selection examination results, and it seems that if allowances could be made for all environmental differences the premature children would on the average do as well in these tests and in the 11+ selection examinations as their controls.

The position of very small premature children needs further mention.

Drillien (1958) reports that children of 3 lb. (1.4 kg.) or less in birth weight are likely to be grossly retarded at both pre-school and school ages. There are few children of really low birth weight in the present study,

TABLE VII.—*Scores and Grammar School Entry of Premature and Control Children Grouped According to Differences in Social and Education Background, Maternal Care, and Interest in School Progress*

Marks for Social and Educational Background, Maternal Care and Interest in School Progress	8-Year Tests			11-Year Tests			Grammar School Places*		
	T Scores		Difference	T Scores		Difference	Premature Children %	Control %	Differences %
	Premature Children	Controls		Premature Children	Control				
Favourable to premature child +3	54·9	48·0	+6·9†	53·2	47·2	+6·0	25·0	0·0	+25·0
Favourable to premature child +2	50·8	50·9	−0·1	51·0	50·6	+0·4	22·2	11·1	+11·1
Favourable to premature child +1	49·0	48·9	+0·1	48·3	48·9	−0·6	12·5	10·0	+ 2·5
Same 0	49·2	49·8	−0·6	47·6	49·3	−1·7	14·8	19·7	−4·9
Favourable to control child −1	48·2	52·2	−4·0†	46·6	52·2	−5·6†	5·7	28·3	−22·6†
Favourable to control child −2	46·8	52·8	−6·0†	45·6	52·3	−6·7†	6·1	32·7	−26·6†
Favourable to control child −3	44·1	54·4	−10·3†	43·8	55·8	−12·0†	0·0	47·6	−47·6†

* Excluding Scotland, areas in England and Wales where there was no 11+ allocation, and all children going to private schools.
† Highly significant.

but the matter is so important that the following figures —scanty though they are—may be of interest.

The relation of birth weight to 11-year-test scores was as follows:

	T score (average of four tests)
-4 lb. (1.8 kg.)	46.4
-4½ lb. (2 kg.)	47.5
-5 lb. (2.3 kg.)	46.4
-5½ lb. (2.5 kg.)	47.5

There were 41 survey children of birth weight 4 lb. (1.8 kg.) or less alive at 8 years. Three of these had gone abroad with their families, 35 were tested at either 8 or 11 years or both, and three were not tested at all for various reasons. The distribution of test scores is shown in Table IX, using 11-year average scores where available or otherwise the 8-year scores:

TABLE IX

Birth Weight	Test Scores (Average of Four Tests at 11 Years)				
	<30	30–39	40–49	50–59	60 and Over
3 lb. (1·4 kg.) or less	0	1	2*	1	0
-3½ lb. (1·6 kg.)	0	2*	4	3	0
-4 lb. (1·8 kg.)	1	7†	6	7	1

* One had 8-year test scores only.
† Two had 8-year test scores only.

Since there is a medical examination for the families of emigrants it may be accepted that the three children going abroad were not grossly defective and there is no evidence in their early histories to the contrary. [The birth weights of these children were 3 lb., 3 lb. 7 oz., and 4 lb.—(1.4, 1.6, and 1.8 kg.).]

The remaining three children were not tested, for the following reasons: 1 (birth weight 4 lb.: 1.8 kg.) E.S.N. attending a special school; 1 (birth weight 3 lb. 3 oz.: 1.44 kg.) now at a secondary modern school, normal early milestones—parents withdrew from survey when he was 4 years old; 1 (birth weight 4 lb.: 1.8 kg.) followed up until 6 years of age. No suggestion of retardation in medical examination by school doctor. Parents are separated and child may have been adopted.

These figures provide no evidence to suggest that these small babies form a special group which is grossly retarded in later life.

In 1946 there was a lack of special facilities for the care of small premature babies, and only the hardiest were likely to survive. It may be that as facilities have improved so have the number of survivors with mental handicaps. The improvement in national survival figures in recent years for babies of low birth weight is small, but in the Simpson Maternity Hospital, Edinburgh, which was the source of Drillien's cases, there appears to have been a substantial increase in the chances of survival of children in the smallest birth-weight groups.

Discussion

The additional material set out in this paper confirms an earlier study which showed that children weighing 5½ lb. (2.5 kg.) or less at birth make low scores in tests of mental ability and school achievement. It further shows that they are rated adversely by their teachers for attitude to work, power of concentration, and discipline in class, and that they fare badly in the 11+ secondary selection examinations. These handicaps, however, are to be attributed to adverse home conditions—in particular to lack of parental care and low educational aspirations—rather than to the effects of low birth weight or cerebral damage associated with it. The problem then reflects the complex social causes of prematurity. It has long been recognized that a high incidence of premature births is associated with failure to use the available antenatal services; it is also associated with poor standards of maternal care and lack of interest in educational progress.

There are compelling reasons for using controls when investigating a relatively rare condition such as prematurity. The cost and labour of observing a whole population in order to obtain information about a small section of it are prohibitive, especially when a longitudinal study is planned. Yet randomly chosen controls are likely to yield misleading comparisons if they differ in social and biological characteristics from the group under study, and we can seldom exclude this possibility.

In choosing controls for the premature children in this study I was in the fortunate position of being able to match for the characteristics that seemed at that time to be of major importance.[8] Unfortunately, as it turns out, they were not the only ones that mattered where educational progress was concerned, and a blind reliance on the method of paired controls would have led to a misleading interpretation of the differences observed. This was realized only after looking at information on social origins, educational background, and parental attitudes that had been collected for the purposes of an entirely separate study of social class and educational opportunity, and *not* for the purposes of the premature study. In other words, if this inquiry had been limited to a study of the mental development of premature children, the information required for a meaningful interpretation of the results would not have been available and incorrect conclusions would have been drawn. This raises a number of interesting questions about the advisability of using controlled comparisons in socio-medical studies. In particular, the fact that a control group is satisfactory at the beginning of a longitudinal inquiry does not mean that it will still be so at the end.

Summary

In a national study of the mental ability and primary-school progress of "premature" children a number of striking handicaps were found, which were later shown to be of environmental origin rather than the result of low birth weight *per se*. The preliminary findings were that premature children: (*a*) made consistently lower scores than their matched controls in eight tests of mental ability and school achievement (four given at 8 years and four at 11 years); (*b*) were the subject of more adverse comment than their controls by their teachers in respect of their attitude to work, power of concentration, and discipline in class; (*c*) were less than half as likely as their controls to gain grammar-school places in the 11+ selection examination.

These differences are largely explained by the fact that premature birth is not only associated with poor living conditions but also, at each social level, with low standards of maternal care and lack of educational interest.

This study casts doubt on the utility of the method of controlled comparisons in socio-medical studies.

I thank the chairman and members of the joint committee for their help and advice; the medical officers of health

[8] The alternative of selecting controls at random could hardly be considered owing to the excess of males, first-born children, etc., among the premature group.

and the health visitors whose generous co-operation made the survey possible; and the mothers in all parts of the country who willingly answered numerous and detailed questions on their children's health. I owe a particular debt of gratitude to Miss Patricia Heneghan, my research assistant.

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SARCOMATOUS CHANGE IN THREE BROTHERS WITH DIAPHYSIAL ACLASIS

BY

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Sarcomatous change is a rare but well-recognized complication of diaphysial aclasis which has never been recorded in two members of the same family. In the family described here, three affected brothers have died of bone sarcomas in the past 25 years.

"Diaphysial aclasis" (Keith, 1920) is the name commonly used on this side of the Atlantic, but there are several synonyms. Ehrenfried (1915) used the term "multiple cartilaginous exostoses," which has become recognized in the United States of America. Jaffe (1943) and Monro and Golding (1951) called the condition "hereditary multiple exostoses." "Multiple exostoses" was used by Abernethy (1830), Virchow (1876), and MacCallum (1940).

Two abnormalities of bone growth characterize diaphysial aclasis. There are osteochondromas, which are more numerous near the epiphyses of the long bones, which grow most actively (Fairbank, 1949, 1951). However, they have been described on all bones, including those of the face and skull, which are preformed in membrane (Honeij, 1920). The exostoses lie between the bone and the periosteum, and show no tendency to erode the parent bone. The ossification of the epiphysial plates of affected bones is disordered. Instead of the normal "row formation" of the epiphysial cells (Dodds, 1930; Johnston and Whillis, 1938), there are masses of cartilage of unusual thickness and patches of ossification. The general appearance is of uncontrolled cartilaginous growth, but without proper preparation for the deposition of cancellous bone. The affected bones fail to grow to their proper length, and the metaphyses are expanded into an irregular cylindrical shape.

The exostoses vary in number from one to 1,000. They appear at any time in childhood or adolescence, and continue to grow until bone-growth ceases at about the age of 20 years. After this they remain static unless malignant change occurs. By the age of 10 years 90% of exostoses have been discovered (Stocks and Barrington, 1925). The unequal shortening of the long bones of the legs causes an affected person to be below average height and to have abnormalities of stance such as kyphoscoliosis, coxa vara, knock-knees, and flat-feet. In middle age severe osteoarthritic changes are frequent in the weight-bearing joints. Bessel Hagen (1891) described the shortening of the distal end of the ulna with a tendency to dislocation of the wrist, which is said to occur in a third of affected people.

Family History

Eight members of this family have lived in Hoddesdon (Numbers 2, 3, 4, 7, 14, 15, 16, 17), and most of their relatives live in the Mons areas of Belgium. I was able to interview 21 blood relatives, and all except two of them allowed me to make a full clinical examination. The oldest living relative (Case 1) denied all knowledge of the family complaint, but she allowed me to palpate her arms and legs. I found exostoses around her knee-joints and osteoarthritic changes in these joints. Five of her seven children had diaphysial aclasis, and three of them have died from bone sarcomas. From the family tree (see next page) it appears that the common female ancestor, who is marked "R," must have carried the gene for diaphysial aclasis. Thirty-two of her direct descendants have been traced; 19 were examined fully; two were interviewed; seven were not seen personally; and four were dead at the time of this survey.

In this family there were seven affected males and six affected females. Ten females and nine males were not affected, but it is likely that some of the younger children will develop exostoses as they grow up. Adults with a very small number of exostoses may have been overlooked, as it was not possible to arrange for *x*-ray films to be taken of all members of the family.

All the affected persons had an affected parent. There was no instance of diaphysial aclasis being inherited through an unaffected parent of either sex. The 13 affected people have had a total of six affected male and four affected female children, as well as five unaffected children of each sex. Exactly half the children of the affected parents have diaphysial aclasis.

It was the "lumps on the bones" which the patients had noticed, and about which they talked. On the whole they had caused very little inconvenience. Only in Case 8 had an exostosis been removed from the lower end of his right radius, at the age of 11 years. The patient's right wrist had been weak since the operation as there was shortening of his radius, which may have been due to damage to the growing epiphysis. In no case was there any symptom from pressure of a benign exostosis on to a joint, tendon, artery, vein, nerve, pelvic outlet, or any other structure. In the nine cases examined the number of exostoses varied from 8 to 78, and were found on most of the bone that had been preformed in cartilage. No exostosis was found on a "membrane-bone." Exostoses were found around the knee-joints of all affected persons. Where an exostosis could be palpated it was usual for *x*-ray examination to reveal several other small nodules. In Case 7, who was 14 years old, the exostoses on her scapulae appeared larger by palpation than on the *x*-ray film. In this case the bulk of the exostoses must have been composed of radiolucent cartilage.

None of the patients had noticed the abnormal bone formation at the metaphyses. The cylindrical section and the abnormal internal structure of the affected metaphyses were easily seen on the *x*-ray films. None of the affected people were above average height, and in four cases (Nos. 2, 7, 10, and 12) one leg was 1 in. (2.5 cm.) or more shorter than the other. In four (Cases 1, 10, 18, and 20) out of seven of the affected people over the age of 40 osteoarthritis of the knees was very troublesome. Case 20 had well-marked coxa vara, knock-knees, and flat-feet, but these may have been due to healed rickets as she had been born near Mons in 1915, and had spent her childhood in this war-stricken area.

Case Histories

Case 2.—In 1956 this man died of chondrosarcoma of the ilium at the age of 54. He had always been aware of