A Family Study of Neonatal and Late-diagnosis Congenital Dislocation of the Hip

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Only during the past 10 years has it been generally appreciated that congenital dislocation of the hip (CDH) can be found in the newborn child if looked for specifically within the first few days of life. Before this, diagnosis was usually made only after the child started to walk. During the past decade there has been at least a fourfold increase in the number of infants attending for treatment. Previous epidemiological and genetic studies have mostly not included the group with neonatal diagnosis. There is, in fact, no certainty that they are both of the same aetiology, and the present survey was undertaken with the object of studying neonatal CDH (diagnosed during the first 4 weeks of life, usually in the first week) and late-diagnosis CDH (diagnosed after the first 4 weeks, usually many months or years later). Only primary congenital dislocation is considered here, not cases associated with arthrogryposis or paralytic dislocation secondary to spina bifida and cerebral palsy.

An attempt has been made to establish the incidence, and to determine the family aggregation of cases, parental age, birth order, pregnancy history, season of birth, and social class. In addition, an investigation relating to familial joint laxity and to acetabular dysplasia in CDH has been carried out, but these clinical findings will only be summarized here and details reported in another paper.

Incidence of Neonatal CDH

It is known that the number of neonatal CDH cases diagnosed depends not only whether the deformity is looked for specifically by a competent examiner, but also on when, during the first week of life, the examination is made. The hip can easily be 'clicked' in and out of joint in the first two or three days of life, and more cases will be found then than in the second three days, when the child's muscles have tightened and the sign can no longer be elicited (Barlow, 1962; Nelson, 1966; Thieme *et al.*, 1968).

Thus, the population incidence of neonatal CDH appears to fluctuate more widely than is the case with more easily observed deformities, though it is probable also that there are wide racial and geographical differences. During the period that specific examination of the neonate has been carried out, figures from England include the following: Barlow (1962) 139 of 9289—15.0/1000 live births (about half these hips were classed as 'unstable', not dislocated); Wilson (1964) 35 of 5700—6.1/1000 live births; Finlay, Maudsley, and Busfield (1967) 60 of 14,594—4.1/1000 live births. In Edinburgh a Congenital Malformations Register was established in 1964, and figures from it for neonatal CDH during the period of this survey are as follows—1965: 69 of 11,080—6.2/1000 live births; 1966: 42 of 10,698—3.9/1000 live births.

These figures must include some patients (though it is impossible to know which ones) who, 10 years ago, would have presented as late-diagnosis CDH as this is a diagnosis now rarely made in the city of Edinburgh, where some 90% of births are institutional and neonatal screening is good.

Incidence of Late-diagnosis CDH

It is unfortunately impossible in retrospect accurately to ascertain this figure, as hospital disease-indexing systems are very incomplete. However, unlike neonatal CDH, this is a well-defined group in which there is invariably deformity and no doubt as to diagnosis. In the decade before 1960 (i.e. before the diagnosis of CDH in the neonatal period became commonplace) it was found that the numbers attending hospital for treatment were approximately one-quarter of the present numbers. Thus, a figure of about 1 per 1000 live births for late-diagnosis CDH is thought to be reasonable. This figure is comparable with those of other surveys which were carried out before neonatal CDH was well established (Record and Edwards (1958) 0.65/1000; McIntosh *et al.* (1954) 1.6/1000).

The current figure for late-diagnosis CDH in the city of Edinburgh has now dropped to 0.13/1000 (G. P. Mitchell, 1970, personal communication). This is accounted for by the occasional child with a 'tight' dislocation who was examined at birth and in whom it was not possible to 'click' the hip in and out of joint. Thus, the dislocation was missed until some months later. It should be noted that late-diagnosis CDH cases continue to come in from the outlying areas which have less adequate facilities for neonatal screening.

Material and Method for Family Study

This survey has been undertaken on the patients from two large orthopaedic clinics, one from the Princess Margaret Rose Orthopaedic Hospital, Edinburgh, and the other from the Royal Hospital for Sick Children, Glasgow. There were 589 index patients (Table I).

TABLE I

		Edinburgh	Glasgow	Total
Neonatal CDH	Male	42	3	45
	Female	140	7	147
	Total	182	10	192
Late-diagnosis CDH	Male	18	34	52
	Female	186	159	345
	Total	204	193	397
	Total	386	203	589

In view of the uncertainty of diagnosis in neonatal CDH, it was considered preferable to include patients only from these orthopaedic clinics with their standardized methods of diagnosis and treatment, rather than attempt a complete survey of the area. It was, however, ascertained from the Congenital Malformations Register for Edinburgh that 75% of the 111 neonatal CDH cases recorded in 1965 and 1966 were included in this survey. The records of the remaining 25% on the Register were checked to ensure they differed in no way from the 75% sample included here. The points noted were the sex ratio, side of deformity, pregnancy, and perinatal history, social class, season of birth, and family history of CDH. The other neonatal cases in the survey were attending the orthopaedic clinics during 1965 and 1966, but were resident outside Edinburgh.

The late-diagnosis CDH patients were all who could be traced from current and old hospital notes dating back to 1932. This cannot be a complete survey of the area, but it is extremely unlikely it is anything but a large random sample.

All 589 index patients were examined either by myself or one of two medically qualified assistants of the former Medical Research Council Research Group on Genetic Problems in Orthopaedic Disease, and an attempt was made to see all first degree relatives either as they attended for a routine clinic visit or by appointment in their own

TABLE II
CDH SURVEY—TRACING OF FIRST DEGREE
RELATIVES
(589 index patients)

	Total No.	Dead	Not Traced or Overseas	Available for Visiting	Refused to Co-operate	Examined
Fathers	589	45	79	465	13	452
Mothers	589	24	25	540	5	535
Brothers	565	65	108	392	9	383
Sisters	549	52	73	424	4	420
Sons	53	—	7	46	—	46
Daughters	67	2	4	61	—	61
Total	2412	188	296	1928	31	1897

homes, whether in Scotland, England, or Wales. Of those available for visiting 98.4% were seen and only 1.6% refused (Table II).

A history was taken relating to CDH and other congenital or developmental anomalies among second and third degree relatives (first cousins only), and, if positive, these individuals were either visited or confirmation of the diagnosis obtained from local medical records. Hearsay evidence only was not accepted.

Examination for excessive joint laxity was carried out in all index patients and all first degree relatives who could be traced. In the acetabular dysplasia investigation, radiographic examination of the hips was carried out in the apparently normal parents of 162 index patients (95 neonatal and 67 late-diagnosis CDH). This part of the survey was continued only until there were adequate numbers for analysis. Control groups from the normal population were also obtained for joint laxity and acetabular dysplasia.

Results

Sex ratio. There was the usual marked preponderance of females in both neonatal and late-diagnosis CDH, the sex ratio being 0.31 in the neonates, and 0.15 in the late-diagnosis group.

Parental consanguinity. There was only one instance of parental consanguinity, the parents being first cousins and having one male neonatal CDH child. The parents and three sibs (two brothers and one sister) were normal.

Familial incidence. The 589 index patients were drawn from 559 families, there being a number of instances of more than one index case in the same family. In 26 cases a double count was used, and in 2 cases a treble count. (Details of kindreds (first degree relatives) are shown in the Appendix.)

Twins. There were five twin pairs in the survey, as follows:

MZ females (neonatal CDH)	—concordant
MZ males (late-diagnosis CDH)	—discordant
DZ females (two pairs, late-diagnosis CDH)	—discordant
DZ { male (neonatal CDH)	—concordant
female (late-diagnosis CDH) }	

TABLE III
PROPORTION OF FIRST DEGREE RELATIVES WITH NEONATAL AND LATE-DIAGNOSIS CDH

Index Patients	Fathers	Mothers	Brothers	Sisters	Sons	Daughters
<i>Neonatal CDH</i>						
Male index (45)	0/45	1/45 (2.2%)	1*/18 (5.6%)	1* + 4/25 (20%)	—	—
Female index (147)	0/147	2/147 (1.4%)	1* + 1/86 (2.3%)	13* + 7/78 (25.6%)	—	—
Total (192)	0/192	3/192 (1.6%)	2* + 1/104 (2.9%)	14* + 11/103 (24.3%)	—	—
<i>Late-diagnosis CDH</i>						
Male index (52)	0/52	1/52 (1.9%)	0/61	1* + 4/53 (9.4%)	0/3	0/4
Female index (345)	1/345 (0.3%)	4/345 (1.2%)	3* + 4/369 (1.9%)	13* + 18/376 (8.2%)	3*/51 (5.9%)	9* + 3/66 (18.2%)
Total (397)	1/397 (0.3%)	5/397 (1.3%)	3* + 4/430 (1.6%)	14* + 22/429 (8.4%)	3*/54 (5.6%)	9* + 3/70 (17.1%)
Grand total (589)	1/589 (0.2%)	8/589 (1.4%)	5* + 5/534 (1.9%)	28* + 33/532 (11.5%)	3*/54 (5.6%)	9* + 3/70 (17.1%)

* Neonatal CDH. All other affected relatives were late-diagnosis CDH.

The diagnosis of mono- or dizygotic twins was made on clinical examination only.

First degree relatives. The proportion of affected first degree relatives is shown in Table III.

It is necessary to distinguish between the affected relatives diagnosed in recent years when many neonatal cases will be included, and those before 1960, which consist of late-diagnosis CDH only. It is noted that the proportion of affected sibs in neonatal CDH (13.5%) is similar to that of affected children in late-diagnosis CDH (12.1%), both these groups being largely post-1960 diagnoses. The figures for parents will relate to late-diagnosis CDH only.

In comparing the familial with the population incidence, the 'expected' figures shown are based on incidence values of 1/1000 and 4/1000 (Table IV). The increase of observed over expected figures is highly significant in each case. There was no significant difference in the proportions of affected

relatives of the male and female index patients. There were many more affected female than male relatives: 14 of 1177 (1.2%) males, compared with 81 of 1191 (6.8%) females. This is to be expected from the known sex ratio of the disorder. These findings are comparable with those in studies from Birmingham (Record and Edwards, 1958) and London (Carter, 1970).

Second and third degree relatives. The proportions of affected second and third degree relatives are given in Tables V and VI. As the information was gained only by history taking (and confirmation from medical records), it is likely that it is less complete for the second degree relatives, many of whom were elderly or dead, than for the third, as these were all first cousins and mostly contemporary with the index patients.

The over-all incidence amongst second degree relatives is 22 of 6664 (0.33%). The same

TABLE IV
CONGENITAL DISLOCATION OF HIP IN FIRST DEGREE RELATIVES

	No.	Affected (Observed)			General Population Incidence Expected Figure Based On:	
		Neonatal CDH in Relatives	Late-diagnosis CDH in Relatives	Total	1/1,000	4/1,000
<i>192 Neonatal CDH</i>						
Parents	384	—	3	3 (0.8%)	0.38	—
Sibs	207	16	12	28 (13.5%)	0.21	0.84
<i>397 Late-diagnosis CDH</i>						
Parents	794	—	6	6 (0.8%)	0.79	—
Sibs	859	17	26	43 (5.0%)	0.86	3.44
Children	124	12	3	15 (12.1%)	0.12	0.48

TABLE V
PROPORTIONS OF AFFECTED 2nd DEGREE RELATIVES WITH NEONATAL AND LATE-DIAGNOSIS CDH

Index Patients	Grand-fathers	Grand-mothers	Uncles	Aunts	Half-brothers	Half-sisters	Grand-sons	Grand-daughters		Nephews	Nieces
<i>Neonatal CDH</i>											
Male index (45)									Brothers' children	—	—
Paternal side	0/45	0/45	0/65	0/62	—	—	—	—	Sisters' children	—	—
Maternal side	0/45	0/45	0/65	0/52	0/2	0/2	—	—			
Female index (147)									Brothers' children	—	—
Paternal side	0/147	0/147	0/232	0/208	0/2	*1/2	—	—	Sisters' children	—	—
Maternal side	0/147	0/147	1/251	2/205	—	0/1	—	—			0/1
<i>Late-diagnosis CDH</i>											
Male index (52)									Brothers' children	0/15	*1/14
Paternal side	0/52	0/52	08/8	0/89	0/4	—	—	—	Sisters' children	0/9	0/10
Maternal side	0/52	0/52	0/101	1/120	0/4	0/2	0/2	0/2			
Female index (345)									Brothers' children	0/88	1/81
Paternal side	0/345	1/345	0/59	2/575	0/13	0/12	—	—	Sisters' children	*1/107	3/98
Maternal side	0/345	0/345	1/54	6/543	0/12	1/12	0/8	0/9			

* Neonatal CDH, all other affected relatives were late-diagnosis CDH.

TABLE VI
PROPORTIONS OF AFFECTED 3rd DEGREE RELATIVES (1st COUSINS ONLY) WITH NEONATAL AND LATE-DIAGNOSIS CDH

Index Patients	Paternal Side				Maternal Side			
	Uncles' Children		Aunts' Children		Uncles' Children		Aunts' Children	
	Male	Female	Male	Female	Male	Female	Male	Female
<i>Neonatal CDH</i>								
Male index (45)	0/44	0/42	0/48	0/43	1*/28	0/14	0/20	0/31
Female index (147)	0/107	0/126	1*/131	1*/135	0/108	1*/105	0/123	1* + 1/127
<i>Late-diagnosis CDH</i>								
Male index (52)	0/58	1/47	0/78	0/76	0/97	1/71	0/149	0/104
Female index (345)	0/476	1* + 1/432	2/483	1* + 1/481	0/364	1* + 1/360	0/477	1* + 1/467

* Neonatal CDH. All other affected relatives had late-diagnosis CDH.

preponderance of affected females, 19 of 3278 (0.58%), is apparent when compared with affected males, 3 of 3386 (0.09%). Among second degree relatives there was an increase ($\times 3$) of the number of affected relatives on the maternal side of the family when compared with the paternal side, but little difference between the numbers of affected brothers' children (1.0%) and sisters' children (1.8%). Among third degree relatives the proportions of affected maternal and paternal were identical (0.3%).

Parental age and birth order. The paternal age in relation to the maternal was noted. The fathers averaged 2.8 years older (mean for whole survey, for male and female index patients separately, and for neonatal and late-diagnosis groups separately). This does not differ significantly from the expected figure for the normal population.

The maternal age and birth order of the neonatal and late-diagnosis groups were compared with the Registrar General's figures for Scotland. A preponderance of first-born children was noted and this was found to be independent of a maternal age

effect. The excess of first-born was highly significant for both neonatal and late-diagnosis groups (Table VII).

Pre- and perinatal factors. These were determined by questioning the mother, where possible, but as in all retrospective history taking the information may not be entirely accurate.

There was no evidence of undue maternal infection during pregnancy. Toxaemia of pregnancy

TABLE VII
BIRTH ORDER OF PATIENTS WITH CDH (EXPRESSED AS NUMBER OF PREVIOUS PREGNANCIES)

<i>Neonatal CDH*</i>	0	1	2 +	Total
Observed	94	54	44	192
Expected	68.1	60.3	63.6	192.0
<i>Late-diagnosis CDH†</i>				
Observed	167	102	115	384
Expected	121.1	120.4	142.6	384.1

* $\chi^2_{(2)} = 16.55$ Highly significant.

† $\chi^2_{(2)} = 25.55$ Highly significant.

occurred in 7% of the mothers of late-diagnosis CDH patients, and in 10% of the mothers of neonatal CDH patients. This latter figure is a little higher than the usually quoted 'expected' figure of 7%. A history of breech presentation or version late in pregnancy was obtained in 17.7% of the neonatal CDH patients (females 15.6%, 18 breeches and 5 versions in 147 births; males 24.4%, 8 breeches and 3 versions in 45 births); and in 17.9% of late-diagnosis CDH patients (females 17.9%, 49 breeches and 9 versions in 324 births; males 20.4%, 10 breeches, no versions in 49 births; no history was available in 18 cases). It was not possible to determine how many were breech with extended legs, which is the presentation thought to be particularly associated with CDH.

The mean birthweight of the neonatal CDH patients was 3448 g. (females 3401 g., males 3492 g.), and of the late-diagnosis CDH patients 3088 g. (females 3129 g., males 3039 g.). The mean for late-diagnosis CDH is exactly the same (3088 g.) as reported by Record and Edwards (1958), and is probably below the expected mean for this area. Unfortunately control figures are not available, and further investigation of both neonatal and late-diagnosis birthweights is needed.

Season of birth. There was a preponderance of CDH births in the two winter quarters of the year (October to March), with a winter:summer ratio of 1.5 for neonatal CDH and 1.3 for late-diagnosis CDH.

Social class. The socio-economic status of the patient's family was classified using the Registrar General's grading based on the father's occupation. No previous survey of late-diagnosis CDH has noted any social class effect, and none was observed in this study. However, the neonatal CDH group showed a significant increase in the proportions of social classes 1 and 2 (i.e. professional and managerial), whereas classes 3, 4, and 5 did not differ from the expected figures. This was apparent in the over-all figures for the whole group of 192 neonatal CDH when 25% were found to be of social class 1 and 2 instead of an expected figure of approximately 15%. This was an unexpected finding and in order to establish it as accurately as possible the social class grading was repeated by a research worker from the Department of Social Medicine, University of Edinburgh, without being aware of the grades previously given. Only minor differences were noted. The social class of those neonates in this survey born only within the city of Edinburgh was then compared with the Registrar General's figures for the city, with approximately the same

result (28% in social classes 1 and 2). Finally, all cases of neonatal CDH recorded in the Edinburgh Congenital Malformations Register for three complete years (1965-1967) were compared with the Registrar General's figures for Edinburgh city (Table VIII), when 29.2% were found to be in social classes 1 and 2 compared with an expected figure of 18.8%.

TABLE VIII
SOCIAL CLASS IN NEONATAL CONGENITAL DISLOCATION OF HIP

	Social Class			
	1 and 2	3	4 and 5	Total
Edinburgh city Neonatal CDH (from Congenital Malformations Register)	45	66	40	151
Normal population (Edinburgh, 1961)	29.3	80.5	41.2	151.0

$$\chi^2_{(2)} = 11.05; 0.01 > p > 0.001$$

In view of this finding, the whole survey (589 patients) was divided by social class and the incidence of CDH in sibs then recorded (Fig. 1). It is clear that there is a social class effect throughout, most obvious when neonatal CDH is included, but apparent to a lesser degree with late-diagnosis CDH also.

Genetic counselling. The risks of CDH to subsequent children in the family calculated on the whole survey (and including both neonatal and late-diagnosis CDH) are as follows: With normal parents, the risk to subsequent sibs is 6% (brothers 1%, sisters 11%); with an affected CDH parent, the

FIG. 1. The figure shows the proportions of affected sibs with neonatal and late-diagnosis CDH, divided by social class, and illustrates the preponderance of CDH in the higher income groups.

risk for a child is 12% (sons 6%, daughters 17%); with an affected CDH parent and one CDH child, the risk to a second child is 36%.

The figures are higher than have been quoted in previous surveys, but include a large number of neonatal cases that in previous years would not have been recognized and would have spontaneously recovered. In view of the social class effect found, 'risk' figures are higher for classes 1 and 2, and lower for 4 and 5. Numbers are only sufficient to calculate for sibs as follows:

Social classes 1 and 2, the risk to sibs is—16.2% (7.5%)

Social class 3, the risk to sibs is—8.9% (5.3%)

Social classes, 4 and 5 the risk to sibs is—3.0% (2.8%)

(The figures in brackets refer to affected sibs with late-diagnosis CDH only.)

Summary of Clinical Investigations*

Acetabular dysplasia. Shallowness of the acetabulum invariably accompanies late-diagnosis CDH, and the question is whether this is a primary growth defect or merely a result of the dislocated head of femur failing to stimulate growth in the socket. Faber (1937) mentioned acetabular dysplasia as a possible factor in CDH. Wilkinson and Carter (1960) measured the 'normal' hip in unilateral CDH cases and found it was frequently dysplastic. In this survey measurements of the acetabulum in the CDH patients themselves were not found to be meaningful, as many children were not fully grown and radiographic measurement of an incompletely ossified acetabulum proved to be unsatisfactory. Measurement of the allegedly 'normal' hip in unilateral cases was also found unsatisfactory, probably because most of these hips had been subjected over a period of years to considerable disturbance by immobilization and plaster fixation during treatment of the dislocated side. It is believed that adequate radiographic measurement of the acetabulum is only possible in adults, and figures for the parents of 162 index patients in this survey were compared with a control series. It was found that the apparently normal parents of the late-diagnosis CDH patients had a significantly shallower acetabulum than the control group. The figures for parents of the neonatal group showed the same trend, but did not reach a significant level (Fig. 2).

Familial joint laxity. There have been previous reports of excessive joint laxity in CDH,

* Details are reported in another paper (Wynne-Davies, 1970).

ACETABULAR DYSPLASIA IN CONTROLS AND CDH PARENTS
C/E ANGLE — AGE CORRECTED SCORES (Both hips)

FIG. 2. Radiographic measurement of the acetabulum was made using the CE angle. This is obtained by joining the central points of the left and right femoral heads (which are spherical); erecting a perpendicular, and drawing a line from the centre to the edge of the acetabulum. The angle between is the CE angle and is a measure of the coverage of the femoral head and of the depth of the acetabulum. The angle varies on left and right sides, and in males and females, also it increases gradually throughout adult life. The histogram shows age-corrected scores for controls, and the parents of neonatal and late-diagnosis CDH patients. The latter have a significantly shallower acetabulum than the controls. (For details see Wynne-Davies, 1970).

thought to be due to an anomaly of oestrogen metabolism (Andrén and Borglin, 1961), or to familial joint laxity (Carter and Wilkinson, 1964a). In a previous study from this centre (Thieme *et al.*, 1968), it was not found possible to confirm an anomaly of oestrogen metabolism, but this current survey did confirm the presence of excessive joint laxity both in the patients and in their first degree relatives (Fig. 3 and 4). This was particularly a feature of the neonatal CDH group. It was also found in the joint laxity control survey of normal children, that under the age of 2 years (but not over) there was a social class effect, with more lax-jointed children in classes 1 and 2 than expected (significant at the 5% level, $\chi^2_{(1)} = 4.1$).

There was further evidence for some connective tissue disturbance in both the patients and their

FIG. 3. This illustrates the amount of joint laxity regarded as excessive: elbows and knees extended beyond 180 degrees, the thumb touching the forearm on flexing the wrist, the fingers parallel to the forearm in extending the wrist and metacarpo-phalangeal joints, and the foot dorsiflexed to 45 degrees or less. If three of the five pairs of joints examined in any one individual showed this degree of laxity it was taken as positive.

FIG. 4. The graph shows the percentage of individuals in each group examined who had excessive joint laxity. It is based on the examination of 192 neonatal and 397 late-diagnosis CDH patients, 1897 of their first degree relatives, and 3354 controls.

relatives, in their associated anomalies. Herniae were particularly common, and there were rather more cases of club-foot and idiopathic scoliosis than expected. Other common congenital malformations occurred no more frequently than would be expected in any random survey.

Discussion and Conclusions

Environmental factors. The findings of this survey confirm others in noting an excess of first-born children, which is independent of a maternal age effect (Record and Edwards, 1958; Woolf, Koehn, and Coleman, 1968). An increased number of breech presentations or versions has also been reported in many other surveys (Muller and Seddon, 1953; Record and Edwards, 1958). It is known that this presentation is particularly common in primiparae (whether the child has CDH or not) and some of the parity effect may thus be accounted for. However, there was still an excess of first-born children even after excluding breech births.

CDH is commoner in the winter months, and again this has been reported in other surveys (Record and Edwards, 1958; Woolf *et al.*, 1968). In many cases this is probably related to the fact that the child is kept more tightly wrapped in the winter and the hips forced into extension. Thus a hip which would reduce itself were the child allowed to kick, is maintained out of joint and then secondary acetabular dysplasia develops. This explanation, however, cannot account for the increase of winter CDH noted by direct examination of the neonate during the first few days of life, and some additional factor must be acting.

The social class effect noted in the neonatal CDH group and in the joint laxity control survey has not previously been described. It could be argued that upper class families look more carefully at their children and thus larger numbers of children with neonatal CDH are diagnosed, but the great majority of Edinburgh births are institutional, and it is on routine examination in the maternity hospitals that the diagnosis is made. Moreover, it is probable in this country that the children of the lower social classes are in fact subjected to more frequent and searching medical examinations than the higher income groups. The effect has not previously been observed because though neonatal CDH is commoner in social classes 1 and 2, it is this particular type that in the past mostly corrected itself and has therefore not been included in earlier surveys. This point has been discussed under 'environmental factors', but it could also have a genetic basis.

Genetic factors.

Family incidence. There were too few twins in this survey for any conclusions to be drawn from the figures, but a large twin survey was reported by Idelberger (1951) in which 10 of 29 monozygotic twins were concordant (35%) and only 3 of 109 dizygotic twins (3%). These figures will certainly relate only to late-diagnosis CDH. Kambara and Sasakawa (1954) reported 27 twin pairs from Japan, in which 15 of 21 'monochorionic' twins were concordant and 3 of 6 'bi-ovular' were concordant. It is not known if this was a complete survey of the area. It is clear that MZ twins are not invariably concordant, but they are so more frequently than DZ twins, indicating a part genetic and part environmental aetiology.

The proportions of affected first degree relatives are significantly in excess of the expected figures for the normal population, though it is noted that the figures for affected parents (0.8%) is low compared with those for sibs and children. This results partly from the absence of neonatal diagnosis in their generation, but probably also from the fact that fewer CDH patients than normal individuals marry. In the past, treatment of this deformity has often been unsatisfactory or not even attempted, and patients were left with deformity and an ugly limp. It was not possible to obtain accurate figures of the total number of married and unmarried CDH women, since it is clearly more difficult to trace those who have married and changed their names, and the survey probably includes an unduly high proportion of single women. There were only eight men in the survey over the age of 20 years (20-27 years), one was married.

There were approximately equal numbers of affected sibs and children (13 and 12%) and though the incidence in parents was less than this, it was still significantly in excess of the expected figure for the general population. The pattern of inheritance on the over-all figures for first degree relatives could therefore be dominance of reduced penetrance, or polygenic.

There were more affected second degree relatives on the maternal than the paternal sides of the family, but this probably just reflects a greater knowledge on the mother's part of events in preceding generations on her own side of the family. No such effect was seen among third degree relatives (first cousins), who were mostly contemporary with the index patients and for whom up-to-date information would more readily be available. The rapid fall in incidence from about 12% affected sibs and children to 1.4% affected nephews and nieces (the second degree relatives for whom information is likely to be

complete) and then to 0.3% affected first cousins (the normal population incidence), indicates polygenic rather than dominant inheritance (Carter, 1969).

Inheritance of acetabular dysplasia. The measurements for acetabular dysplasia were normally distributed and it is probable (as implied by Record and Edwards, 1958) that the configuration of the acetabulum is inherited as a multiple gene system. That being so, one would expect that the more severe the degree of dysplasia in the parents, the more relatives would be affected with CDH. This did prove to be the case, and figures for affected sibs, divided according to the severity of acetabular dysplasia in parents, are shown in Table IX. It is thought, therefore, that acetabular dysplasia must be a primary genetic disorder of polygenic inheritance.

TABLE IX

ACETABULAR DYSPLASIA
(Sibs with CDH grouped according to severity of acetabular dysplasia or frank dislocation in parent)

	Mild One Parent 1SD From Mean	Moderate One Parent 2SD From Mean or Two Parents 1SD Or More From Mean	Severe 1 Parent Dislocated
	42 Index Cases	42 Index Cases	9 Index Cases
Affected sibs	4/47 (8.5%)	12/64 (18.8%)	4/11 (36.4%)

It was interesting to note on comparing the group in which parents had acetabular dysplasia (84) with that in which the parents' x-rays were normal (78), that the former had lost certain 'environmental' aspects of CDH. There was no excess of first-born children, and no excess of winter births. Other differences showed in the sex ratio which was now 0.5 and the much greater frequency of bilateral cases (74% compared with 32%).

Inheritance of joint laxity. The measurement of excessive joint laxity is not so precise as measurement of the acetabulum, but since lax joints were demonstrable not only in the index patients, but in parents, sibs, and children in significantly greater numbers than in a control survey, it is thought that this must be one aetiological factor in CDH. It is difficult to establish the exact numbers of affected families because joint laxity diminishes with age. It was often associated with other connective tissue defects, such as herniae (cf. Marfan's syndrome of dominant inheritance), but the age of appearance of these is very variable. However, in this survey a connective tissue disorder did occur in a high proportion of both sexes and all generations, and the inheritance

may well be dominant. There have been families reported in the literature in which excessive joint laxity appears to be of clear dominant inheritance (Carter and Sweetnam, 1960).

Thus, it is probable that there are two independent gene systems in CDH (as suggested by Carter and Wilkinson, 1964b). One concerns the development of the acetabulum and is a polygenic system. In the past this has contributed to a high proportion of late-diagnosis CDH. The other concerns the laxity of the capsule round the hip joint (as well as the capsules of other joints), contributes to the great majority of neonatal CDH, and is likely to be of dominant inheritance. In the past many of these cases corrected themselves and went unnoticed. Those that remained unreduced would have developed secondary acetabular dysplasia and in due course presented as late-diagnosis CDH. Any group of neonatal CDH patients will contain both aetiologies, though a preponderance (perhaps 80%) of the joint laxity type, and any group of late-diagnosis CDH patients will contain both aetiologies, but a preponderance of the primary acetabular dysplasia type.

It must also be noted that these traits are quite common in the general population, and thus some patients will have both defects. It is, unfortunately, not possible to be sure how many, because the joint laxity group cannot be clearly defined and acetabular dysplasia can only be measured with certainty in the parents. However, in the few families in which it was clear that both defects were present, there appeared to be an increased risk of CDH developing.

Falconer (1965) has developed a method for estimating the relative importance of heredity and environment in quasi-continuous traits, using data consisting of the incidence of the disorder in first degree relatives and in the general population. The correlation between relatives in respect of 'liability' to the disorder can be calculated, and from this the heritability can be estimated. It seems clear that there are two independent gene systems in CDH, though it is not always possible (clinically) to separate them. However, a preponderance of late-diagnosis CDH appears to be of the acetabular dysplasia type. Thus, assuming a population incidence of 1 per 1,000, and noting the proportion of affected relatives to be 64 of 1,777 (3.6%), the heritability is found to be nearly 80%. A preponderance of neonatal CDH appears to be of the joint laxity type. Here, the population incidence is taken as 4 per 1000, the proportion of affected sibs 28 of 207 (13.5%), and the heritability is found to exceed 100%, suggesting that single gene of dominant effect is involved in this case. However,

these figures can only be tentative, since the two systems may act separately or together, and cannot always be identified in the individual patient.

Summary

A family study of 589 patients with congenital dislocation of the hip has been carried out, separating those in whom diagnosis was made in the neonatal period and those with a later diagnosis.

The survey confirms others in finding a family aggregation of cases, an increased number of breech births, a preponderance of first-born children and of children born in the winter months.

There is a significant increase of patients with neonatal CDH in social classes 1 and 2 (the higher income groups).

Evidence is presented indicating that two gene systems are acting: one relates to dysplasia of the acetabulum and is polygenic, and the other to laxity of the capsule round the hip joint, and is probably dominant.

Both traits may be present separately or together, but there is a preponderance of the joint laxity type in neonatal CDH and of the acetabular dysplasia type in late-diagnosis CDH.

I am much indebted to Mr. G. P. Mitchell, Princess Margaret Rose Orthopaedic Hospital, Edinburgh, and Mr. N. J. Blockey, Royal Hospital for Sick Children, Glasgow, for permission to examine their patients and for access to case records. Acknowledgement and thanks are also due to Miss P. Dugard of the Department of Statistics, University of Edinburgh, to Dr. Norman Dean, formerly in charge of the Edinburgh Register of Congenital Malformations, and to Dr. H. Cameron and Dr. D. Smith for carrying out much of the family visiting. Fig. 2, 3, and 4 are published by permission of the publishers of the *Journal of Bone and Joint Surgery*.

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Appendix

Edinburgh/Glasgow CDH Survey

M, male; F, female (italics indicate patients with CDH); n, neonatal diagnosis CDH; l, late diagnosis CDH; m, miscarriage; sb, stillbirth
 *, index patient; [], twins.

e.g. *F1, index, female, late diagnosis.

Serial No.	Sibship	Date of Birth	
		Father	Mother
1 } 2 } 3 } 4 } 5 } 6 } 7 } 8 } 9 } 10 } 11 } 12 } 13 } 14 } 15 } 16 } 17 } 18 } 19 } 20 } 21 } 22 } 23 } 24 } 25 } 26 } 27 } 28 } 29 } 30 } 31 } 32 } 33 } 34 } 35 } 36 } 37 } 38 } 39 } 40 } 41 } 42 } 43 } 44 } 45 } 46 } 47 } 48 } 157 } 49 } 50 } 51 } 52 } 53 } 54 } 55 } 56 } 57 } 58 } 59 } 60 } 61 } 62 } 63 } 64 } 65 } 66 } 67 } 68 } 69 } 70 } 71 } 72 } 73 } 74 }	M 11/61; [*Fn *Fn] 10/64 M 6/57; M 6/59; *Fn 12/64 *Fn 10/64 *Fn 1/65 *Fn 11/64 *F1 5/57; F 5/54; m -/55 F 8/63; *Fn 10/64 M 9/61; M 12/62; *Fn 8/64 *F1 10/60; *Fn 7/64 *Mn 11/62; *Fn 11/64 M 2/59; *Fn 8/63 F 5/62; *Fn 1/64 *Fn 12/64 *F1 12/53 M 10/46; m 6/48; *F1 1/51; M 1/56 m -/57; m -/58; M 4/59; *Fn 3/64 *M1 1/63; *Fn 10/64 *F1 12/58; F 8/62 F 11/42; F 5/50; M 11/54; m -/57; *M1 10/59 *F1 10/55; F 4/58 *F1 1/59; F 12/61 F 6/62; *Fn 1/65 m 10/63; *Fn 1/65 *Fn 9/62; *Fn 12/64; M 12/66 F 2/63; m -/63; *Mn 9/64; m -/65; F 1/67 F 1/64; *Mn 12/64 *Fn 1/65 F 5/55; *F1 7/56; M 3/60; M 7/62 *Mn 1/65 *Mn 3/65 *Mn 6/63; *Mn 3/65 m -/50; M 11/51; *F1 8/53 *Fn 4/65 F 9/58; *Fn 1/63 F 12/56; M 12/60; *F1 9/62; F 3/63 F 11/45; F 9/47; M 9/49; m -/54; m -/58; *Fn 5/63 *Fn 2/64 F 2/60; M 1/63; *M n 5/65 *Mn 7/64 F 5/62; *Fn 5/65 *F1 10/61; M 10/64 *F1 4/58; M 7/60; *F1 12/65 m 2/64; *Fn 6/65 *F1 12/58; *F1 3/65 *Fn 5/65; M 10/66 *F1 9/63 F 11/51; F 2/54; M 12/59; *F1 10/61; F 1/64 M 4/58; F 9/60; *Fn 1/65 *F1 11/62 F 1/54; F 9/56; M 8/61; m -/63; *Fn 1/64 m 8/62; *Mn 8/65 M 2/61; *F1 1/62; M 4/63 F 9/53; *F1 7/56 M 3/62; *Fn 8/65 M 3/61; *Fn 2/62; *Fn 8/65 *Fn 8/59; M 9/60; F 8/63 *Fn 3/65 (See 297) *Fn 6/63 *Fn 8/65 *Fn 6/65 *F1 12/63 *Fn 10/62; *F1 7/65 *Mn 8/65 *Fn 6/65 M 4/60; F 7/61; *Mn 6/65	1930 1926 1929 NK 1934 1926 1936 1937 1935 1927 1920 1927 1946 1916 1906 1931 1939 1931 1922 1926 1933 1931 1933 1942 1936 1944 1938 1932 1937 1939 1932 1926 1936 1928 1928 1925 1921 1936 1933 1936 1937 1934 1932 1934 1934 1943 1943 1929 1935 1927 1933 1938 1937 1918 1927 1939 1936 1938 1945 1929 1935 1940 1943 1934	1938 1935 1933 1941 1936 1926 1942 1941 1937 1926 1929 1929 1943 1919 1914 1930 1940 1928 1921 1931 1937 1936 1940 1943 1940 1946 1939 1931 1937 1937 1939 1939 1928 1938 1929 1931 1938 1939 1935 1942 1939 1935 1938 1939 1939 1946 1944 1930 1940 1922 1935 1938 1938 1947 1938 1928 1942 1947 1933

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
75	*Fn 9/65	NK	1946
76	F 3/64; *Mn 6/65	1936	1937
77	M 7/59; F 10/61; *Fn 3/65	1925	1929
78	M 10/48; m -/49; F 11/51; F 10/52; F 6/54; M 3/56; M 10/58; M 11/59; *F1 12/61	1919	1929
79	*M1 9/60; M 10/62	1937	1939
80	M 11/49; M 11/52; F 6/54; M 3/57; M 9/61; *Fn 6/65	1912	1926
81	m -/60; *Fn 5/62; M 6/63	1935	1934
82	*Fn 9/65	1937	1944
83	M 2/62; F 4/63; *Fn 10/65	1932	1936
84	*Fn 7/65	1940	1938
85	M 6/54; *M1 12/62	1927	1930
86	*F1 4/63; F 6/65	1912	1932
87	F 10/61; *F1 2/64	1938	1937
88	M 8/57; M 3/62; *Fn 6/65	1926	1936
89	m 10/63; m 3/64; *Mn 10/65	1937	1936
90	*Fn 9/65	1942	1944
91	F 9/59; *F1 3/61; *Fn 4/63; *Fn 7/65	1927	1932
92			
93			
94	M 9/64; *Fn 9/65	1940	1945
95	M 11/64; *Fn 10/65	1937	1938
96	*F1 6/57; M 10/58; *F1 12/60; M 5/63; F 7/65	1935	1930
97			
98	M 9/62; m -/64; *Fn 8/65	1936	1935
99	M 6/62; M 8/63; F 10/64; *Mn 11/65	1936	1941
100	M 12/55; M 7/57; F 4/59; *Fn 9/65	1907	1927
101	M 5/61; *F1 4/63	1926	1934
102	*Fn 6/65	1934	1930
103	*F1 10/63; M 11/65	1937	1938
104	F 10/62; *Fn 5/65	1932	1938
105	M 5/62; *F1 3/64	1932	1936
106	F 6/60; *F1 6/65	1933	1934
107	M 12/57; *Fn 11/65	1925	1932
108	M 1/61; *F1 10/64	1931	1932
109	*Fn 6/63; F 7/65	1942	1946
110	*Fn 7/65	1935	1935
111	*Fn 6/64; M 9/66	1936	1933
112	M 10/62; *Mn 11/65	1935	1942
113	*F1 3/65; M 6/66	1935	1937
114	*F1 10/60; *Mn 3/64	1934	1938
115			
116	m 2/62; F 11/62; *Fn 10/65	1935	1943
117	*Fn 11/65	1935	1934
118	F 1/63; *Fn 4/65	1929	1939
119	*Mn 8/65	1938	1939
120	F -/53; M 10/54; m -/55; F 7/57; *F1 12/58	1926	1929
121	F 11/58; M 3/61; *Fn 9/65	1935	1937
122	F 1/55; M 6/57; M 7/59; *Fn 9/62; F 4/67	1927	1932
123	*Fn 8/65	1938	1938
124	*F1 6/64	1940	1942
125	M 12/63; *Fn 9/65	1935	1938
126	F 7/57; *M1 1/60; F 4/62	1926	1930
127	*Mn 10/65	1925	1928
128	M 7/61; M 1/63; M 3/64; *Fn 8/65	1934	1937
129	*Mn 12/65	1945	1946
130	sbM -/56; M 7/57; *F1 10/59	1920	1920
131	M 10/64; *Fn 11/65	1940	1946
132	*F1 10/63	1942	1942
133	F 5/57; M 4/60; *Fn 10/64	1927	1926
134	*Mn 11/65	1940	1942
135	M 3/58; F 2/60; *F1 10/62	1924	1926
136	*Fn 6/65; F 11/66	1938	1940
137	*Fn 11/65	1933	1947
138	M 9/62; *Fn 3/65	1938	1941
139	*Fn 8/65	1940	1937
140	F 4/64; *Fn 12/65	1942	1943
141	*Mn 11/65	1924	1941
142	*F1 2/65; *Fn 1/66	1936	1940
143			
144	*Fn 2/64	1925	1930
145	*Fn 11/65	1938	1944
146	*F1 4/58; F 5/62	1932	1934
147	M 2/56; sbM -/58; F 1/59; F 1/62; *Fn 6/65	1944	1943
148	M 5/43; M 12/44; M 2/46; *F1 7/49; M 6/55; *Fn 9/61	1929	1932
149			
150	*F1 1/49; M 10/52	1917	1921
151	(See 142)	1924	1926
152	M 9/64; *Fn 1/66	1939	1938
153	*Fn 12/65	1934	1934
154	*Fn 2/66	1937	1938
155	m -/63; M 4/64; *Fn 12/65	1940	1940
156	*Fn 1/66	1941	1943

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
157	(See 48)		
158	F 8/49; *F1 10/56	1919	1916
159	*F1 10/61; F 10/63; *F1 2/65	1936	1933
160	F 1/44; M 12/45; M 4/48; *F1 1/56	1922	1923
161	m -/56; M 5/58; F 10/59; F 12/62; *F1 9/64	1933	1933
162	*Mn 1/66	1935	1935
163	F 2/50; F 6/51; m -/52; M 2/54; F 1/58; m -/59; m -/61; M 4/63; *Mn 7/64	1916	1927
164	*Fn 10/64	1933	1939
165	*Fn 2/66	1930	1939
166	m 4/65; *Fn 3/66	1934	1939
167	*Mn 3/66	1941	1943
168	M 6/63; m 11/64; *Fn 10/65	1937	1937
169	(See 159)		
170	*Fn 12/65	1933	1933
171	F 7/62; *Fn 11/65	1938	1944
172	*F1 9/58	1916	1930
173	*Mn 2/66	1941	1943
174	F 5/64; *Fn 2/66	1944	1945
175	M 1/60; M 7/61; M 8/63; *Fn 11/65	1939	1941
176	m 9/64; *Fn 1/66	1940	1946
177	M 7/50; sbF -/53; F 4/54; M 10/61; *Fn 2/66	1931	1932
178	*Mn 12/65	1925	1924
179	F 9/53; F 1/56; *F1 12/57; m 6/58; m 9/59	1923	1933
180	M 10/51; F 6/53; M 7/56; *Mn 1/63	1929	1930
181	F 7/57; M 12/58; *Mn 8/62; F 2/64	1927	1928
182	M 10/40; F 10/41; M 11/42; F 7/46; M 2/50; *Fn 10/62	1909	1920
183	*Fn 2/66	1935	1939
184	*Fn 2/66	1942	1942
185	m 6/60; M 9/61; M 1/63; *Fn 10/65	1940	1941
186	M 10/53; m -/56; M 10/57; M 1/61; *Fn 5/63	1931	1932
187	*F1 1/46; F 5/51	1920	1915
188	*Fn 1/63	1929	1926
189	*Fn 3/66	1922	1938
190	*Fn 2/66	1940	1942
191	F 7/52; F 9/54; M 12/58; *F1 7/60; M 8/61; F 1/63	1930	1932
192	F 9/60; *F1 4/63	1940	1940
193	F 6/51; [M; F]; 11/54; M 7/57; *F1 11/59	1927	1927
194	*F1 2/63	1940	1938
195	*Fn 3/66	1937	1939
196	*Fn 2/66	1938	1938
197	M 2/49; M 2/51; F 4/53; *F1 9/56	1917	1922
198	[*Mn 1/66; *F1 1/66]	1940	1942
199			
200	F 3/55; M 5/58; F 2/63; *Fn 11/65	1931	1932
201	F 7/59; m 1/60; F 7/62; *Mn 7/63	1935	1938
202	M 6/51; [M; M]; 4/54; M 8/61; *Fn 2/63	1929	1928
203	M 9/56; F 2/59; F 2/60; F 4/64; *Fn 11/65	1919	1928
204	m 3/64; F 3/65; *Fn 3/66	1938	1944
205	*Fn 3/66	1936	1943
206	m -/60; F 1/61; *Fn 3/66	1925	1927
207	m 7/63; F 4/64; *Mn 3/66	1935	1937
208	*Fn 12/65	1930	1935
209	*Fn 7/65	1937	1939
210	M 9/61; M 9/63; F 12/64; *Fn 3/66	1936	1936
211	*Fn 3/66	1944	1945
212	M 12/57; *F1 4/61	1933	1935
213	F 7/21; *F1 11/23; F 4/30; M 10/37	1893	1893
214	*F1 3/38	1905	1915
215	F 5/49; M 12/51; M 6/53; F 5/54; M 2/56; F 1/59; F 4/60; M 3/62; Fsb 8/64; *Fn 4/66	1927	1928
216	*F1 3/25; M 7/27; M 12/28	1902	1902
217	Fsb -/56; *F1 5/58; F 5/60; M 10/61; F 5/64	1916	1926
218	M 2/43; M 10/47; *F1 1/48; m -/5; F 3/53	1914	1913
219	F 10/55; M 4/57; *F1 1/59	1930	1934
220	M 6/52; *F1 9/54	1929	1929
221	*F1 2/52; Fsb 11/56; M 6/58	1926	1927
222	M 1/56; *F1 5/57; M 1/64	1929	1932
223	*Fn 4/66	1940	1942
224	M 4/60; M 12/62; *Fn 3/66	1927	1934
225	*Fn 3/66	1943	1945
226	M 1/65; m 5/65; *Fn 4/66	1945	1947
227	F -/25; M 12/26; F 5/28; F 10/29; *F1 12/30; M -/31; M -/32; m -/34; M -/37; F -/39; M -/43	1907	1908
228	F 11/59; *Fn 4/62; F 7/63	1927	1933
229	M 3/10; M 3/12; M 1/14; *F1 2/15	1884	1889
230	*F1 4/16; F 12/20; M 6/24; M 11/27	1889	1886
231	F 6/11; *F1 10/13; M 2/17; M 2/16	1864	1885
232	*F1 4/43	1909	1912
233	*F1 3/57	1929	1930
234	M 3/65; *Fn 3/66	1929	1932
235	F 10/62; *M1 12/63	1940	1942
236	M 10/41; *F1 4/47; F 12/51; M 1/54	1912	1918
237	*Mn 7/62	1928	1930
238	*F1 9/60	1924	1927

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
239	F 5/59; [F; *F1] 3/61; M 12/62	1928	1933
240	*Fn 10/66	1938	1941
241	F 3/57; F1 6/58; *F1 11/61	1923	1928
242	*F1 3/64	1931	1938
243	F 3/51; *F1 2/57	1925	1924
244	*F1 8/57	1929	1932
245	*F1 6/42	1921	1922
246	*F1 8/48; F 9/50; F1 3/53	1922	1922
247	*F1 11/48; M 12/52; F 1/55; F 2/61	1924	1924
248	*F1 12/26; F -/-; M -/-; M -/-	1897	1897
249	m -/47; *F1 9/48; M 6/51	1927	*1926 (No. 248)
250	*F1 2/51	1921	1919
251	*F1 9/48; M 6/51	1923	1926
252	F 9/50; F -/54; *F1 12/57	1921	1926
253	*F1 6/32; M -/-; M; M; M; M 6/40; M 8/44; M 7/46	1901	1903
254	*Fn 10/64	1928	1925
255	M 1/64; M 7/65; *Fn 7/66	1934	1938
256	m -/-; m -/-; *Fn 9/62; M 6/65	1930	1933
257	*F1 8/63	NK	1940
258	F 12/64; *Fn 2/66	1943	1945
259	M 9/53; m -/55; M 9/57; *F1 5/59; M 5/61	1926	1926
260	M 2/30; m -/31; F 10/31; *F1 12/40	1901	1897
261	M 11/21; *F1 4/27; M 9/29; F 3/33	1901	1904
262	F 11/48; M 2/50; F 4/51; *F1 8/52; F 8/54; M 2/56; Fsb 5/62; M 3/64	1930	1928
263	*Mn 6/66	1925	1930
264	*Fn 5/66	1946	1948
265	*F1 9/54; F 12/57; F 8/61	1929	1931
266	*Fn 11/63	1939	1941
267	m -/59; m -/59; m -/60; *F1 3/65	1929	1927
268	*F1 3/45	1917	1920
269	F 11/56; F 8/58; M 3/62; *Mn 11/63	1927	1929
270	*F1 1/52; F 11/56; M 2/59; M 9/61	1925	1927
271	*Fn 9/62; M 8/65	1933	1937
272	M -/1899; F -/-; F -/00; *F1 3/02; F -/03; M -/05; F -/07; F -/09; M -/11; M -/13; M -/15	NK	NK
273	F1 12/63; *Fn 5/66	1938	1940
274	Msb -/62; *Fn 3/63; F 8/65	1940	1941
275	M 11/27; *F1 12/28; M -/32; M 1/35	1901	1903
276	m -/59; *Mn 11/66	1938	1937
277	*Mn 7/63; *F1 5/66	1935	1936
278		1938	1942
279	*Fn 8/66	1938	1945
280	*Mn 6/66	1923	1921
281	M 3/49; *F1 1/51	1908	1909
282	m -/40; F 6/41; F 3/43; *F1 1/50	1879	1893
283	M 3/18; F -/23; *F1 3/27; F 5/32	1907	1905
284	*F1 3/37	1916	1918
285	*F1 1/46; M 9/50	1915	1924
286	F 12/46; *F1 4/50; F 10/52; F 1/57	NK	NK
287	M 6/06; M 2/08; M 3/10; M -/12; M -/15; *F1 2/16; Fsb -/-	1927	1931
288	*F1 12/45; Fn 1/59; *F1 11/61	1930	1933
290	M 4/53; F 7/55; M 4/58; M 1/60; *F1 7/62 (See 288)	1924	1925
289		1932	1938
291	M 2/46; F 3/48; *F1 2/51; F 4/56; M 2/59	1914	1916
292	F 7/63; *Fn 1/66	1921	1922
293	M 10/34; F 1/37; F -/-; *F1 10/41	1937	1937
294	F 10/43; *M1 5/47; M 11/53	1938	1943
295	m 12/64; *Mn 9/66	1881	1883
296	M 10/62; *Fn 9/66	1906	1915
297	*F1 5/35; M -/-; M -/-; F -/-; F -/-	1946	1943
298	*M1 4/41; m -/44; M 3/48; M 2/52	1935	1936
299	*Mn 11/66	1944	1946
300	F 7/65; *Fn 11/66	1941	1945
301	*Mn 12/66	1942	1943
302	*Fn 12/66	1938	1942
303	*Fn 12/66	1944	1941
304	*Fn 12/66	1939	1937
305	*Mn 12/66	1940	1939
306	*F1 4/65	1932	1935
307	F 10/61; *F1 12/63	1929	1930
308	M 8/54; M 5/55; M 10/57; F 8/60; *F1 12/63	1933	1936
309	M 1/54; M 2/56; F 1/59; *F1 4/63	1938	1935
310	F 8/57; F 12/58; F 1/60; M 3/61; m 5/62; *F1 5/63; M 3/65	1929	1930
311	F 7/58; *F1 9/60; F 10/62	1933	1936
312	*F1 9/63	1938	1935
313	*F1 9/63	1929	1936
314	*F1 1/62; M 2/63; M 5/64	1937	1942
315	F 5/60; *F1 9/63	1935	1939
316	*M1 4/62; F 4/64	1938	1937
317	*F1 2/64	1934	1939
318	M 1/65; *F1 8/62	1939	1940
319	*F1 8/56; M 6/61	1941	1941
		1924	1926

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
320	*F1 5/51; M 7/56	1926	1926
321	M 5/53; M 8/52; M 9/55; F 10/56; *F1 5/58; F 10/60; M 8/61; m -/62; M -/63; M 9/65	1931	1932
322	F 8/60; *F1 9/61; M 12/64	1934	1936
323	M 2/52; F 1/53; *F1 7/61	1927	1929
324	F 5/62; *F1 8/63; F 3/65	1940	1940
325	F 4/57; F 3/59; M 4/61; F -/62; *M1 2/63	1936	1936
326	*F1 11/60	1927	1924
327	F 3/52; F 5/53; m -/60; *M1 4/61	1930	1928
328	M 9/51; Fsb -/52; F -/-; M 1/56; M 3/58; *Mn 3/63	1920	1927
329	*F1 5/60; F 5/65	1930	1936
330	F 11/56; M 4/58; F 12/61; *F1 2/62; F 8/63	1928	1932
331	*F1 9/62	1933	1939
332	M 8/57; *M1 10/58; M 10/59; M 8/61; F 4/63; M 11/64	1932	1939
333	F 3/52; M 6/54; F 4/56; M 8/59; *F1 6/61	1926	1930
334	M 7/60; *F1 7/61	1935	1940
335	F 11/48; F 3/49; M 12/52; F 9/55; *F1 2/61	1925	1928
336	*F1 2/56; Fsb -/59; F 9/61; F 4/65	1931	1932
337	*F1 12/63; F 12/64	1938	1939
338	M 7/53; F 7/54; Msb 6/55; M 8/56; Msb 8/57; F 11/59; *M1 2/61; F 10/62	1904	1928
339	*F1 3/53; M 4/54; M 3/56; M 7/57; F 10/59; m -/61; F 6/62; F 4/65	1923	1930
340	*F1 1/59; *F1 2/62	1931	1933
341	F 8/62; *F1 8/63	1936	1942
342	*F1 7/64	1942	1944
343	M 1/55; *M1 1/60; F 2/62; M 8/64	1929	1931
344	*F1 6/58; M 5/60	1928	1934
345	M 5/57; M 6/58; m 8/52; *M1 12/59; M 4/61; F 9/64	1935	1935
346	*F1 5/54; Msb -/56; F 1/59; F 7/59	1924	1931
347	M 1/51; F 3/54; *F1 12/60	1921	1929
348	F 5/52; *F1 12/53; F 10/56; m -/57; M 3/59; M 9/64	1926	1929
349	M 11/56; *F1 3/58; F 6/62	1935	1939
350	M 8/53; F 8/55; *M1 9/57; F 4/65	1926	1928
351	*F1 1/61; m -/63; M 12/64	1927	1929
352	M 5/55; *F1 12/56; M 8/58; F 4/61	1930	1932
353	*F1 9/57	1912	1926
354	*F1 5/60; F 3/62; m -/63; F 8/64	1933	1932
355	M 12/57; *F1 6/60	1931	1934
356	*Fn 3/60; *Fn 4/63	1935	1936
357	*F1 3/60	1932	1939
358	*F1 4/63	1932	1935
359	M 3/56; F 5/57; *F1 7/61	1931	1931
360	m -/54; *F1 7/56; F 8/59; M 12/60	1932	1932
361	M 4/54; *F1 10/56; F 1/60	1931	1933
362	*F1 10/60; m -/63; F 4/65	1926	1936
363	F 8/49; F 11/50; *M1 6/53; M 7/63	1926	1927
364	*F1 9/61; F 10/63	1938	1939
365	*F1 1/55	1926	1926
366	M 4/37; Msb -/38; M -/39; F -/42; *F1 4/53	1915	1915
367	F 10/44; m -/45; m -/46; m -/47; F 2/48; M 4/50; M 10/51; *F1 12/54; M 4/56; M 6/58	1915	1915
368	*F1 8/58; F 3/62	1932	1934
369	*M1 5/59; M 9/62	1924	1925
370	M -/43; F -/48; F -/55; *F1 2/57; F 3/59; M 8/62	1921	1922
371	*F1 10/50; m -/54	1919	1921
372	*F1 6/51	1920	1920
373	F 12/48; F 11/49; M 7/51; M 10/53; F 3/55; M 8/60; M 8/61; *F1 3/64	1922	1928
374	*F1 9/47; M 3/55	1925	1927
375	F 7/54; *F1 5/58	1928	1927
376	*F1 9/60; F 10/62; F 7/64	1938	1941
377	F 3/58; F 4/59; M 5/60; *M1 3/61; M 8/62; M 3/65	1929	1934
378	M 1/48; F 10/49; *F1 12/52; F 2/54; m -/57; M -/58	1925	1921
379	F 7/55; *F1 1/58	1929	1927
380	M 5/62; M 6/63; *Mn 11/64; F 11/65	1937	1940
381	*F1 8/35; M -/40; M -/46	1906	1906
382	M 6/46; F 10/48; *F1 10/49	NK	1921
383	F 9/46; M 9/48; M 7/49; M 9/51; *F1 3/53; F 10/55; F 12/57; F 3/60; M 10/61; M 2/66	1924	1925
384	F 11/54; M -/-; M 1/57; M 10/58; M 10/59; F 6/61; *F1 2/63; m -/64; m -/65	1928	1929
385	F 4/62; *Fn 5/65	1938	1940
386	Msb -/53; Msb -/54; M 4/57; *F1 1/59; M 1/61; M 1/62; F 5/65	1930	1930
387	Fsb 5/61; *M1 5/62; M 3/64	1942	1943
388	*M1 11/58; F 10/59; M 4/62; M 4/63	1935	1936
389	*F1 8/51	1932	1932
390	F 4/48; M 5/50; *M1 10/56; M 5/59; M 5/63	1931	1928
391	M 11/47; F 4/49; *F1 2/54	1923	1922
392	M 4/52; F 5/54; m -/56; M 9/59; *F1 2/61; m -/63; F 10/64	1926	1930
393	F 4/47; F 2/49; *F1 12/52; F 6/55	1917	1916
394	M 6/49; *M1 12/52	1919	1921
395	F 5/52; *F1 5/56	1922	1924
396	M 4/59; *F1 9/60; F 5/64	1930	1930
397	*F1 4/55; F 6/56; F 1/63; F 3/64	1930	1931
398	*F1 2/55; F 2/58	1928	1929
399	M 7/55; *F1 4/58	1921	1926
400	*F1 2/62; F 2/65	1939	1938
401			
402			

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
403	*F1 5/57	1921	1920
404	*F1 8/55	1921	1920
405	F 12/53; *M1 8/58	1928	1930
406	*F1 5/58; M 4/60; M 2/62; M 7/63	1933	1936
407	M 6/58; *M1 10/62	1933	1935
408	F1 3/42; F 9/46; F 1/48; [F; F;] 12/49; M 5/52; *M1 10/57	1913	1915
409	F 7/43; M 9/46; F 1/51; *F1 1/53; M 11/54	1916	1921
410	*F1 6/53	1922	1924
411	M 12/57; *F1 10/60; [Msb; Fsb;] -/61; F 12/62; M 2/64	1928	1935
412	*M1 11/60	1935	1938
413	M 5/57; M 10/58; F 2/61; *F1 10/62; Fn 6/66	1925	1932
414	F 4/53; M 7/54; *F1 2/56; M 3/57; M 5/58; F 8/60; M 8/61; M 10/63; F 11/64	1931	1931
415	*F1 11/50; M 10/52; M 10/54; *F1 9/56; m -/58; m -/58	1925	1929
416	M 4/55; F 5/56; *F1 6/57; M 8/58; M 9/62	1926	1924
417	F 6/59; *F1 4/63; M 6/65	1933	1937
418	M 2/54; F 2/55; *F1 5/57; F 1/59	1929	1931
420	*F1 10/51; [M; F] 2/53; F 12/56	1920	1922
421	m -/55; F 11/57; m -/55; F 3/61; M 11/62; *F1 8/63	1931	1935
422	Msb 12/50; F 12/51; *F1 1/53; M 4/54; F 12/57; m -/60; m -/62	1927	1931
423	M 8/56; M 10/57; *F1 4/60; F 8/61; F 3/63; F 5/65	1929	1930
424	M 4/52; m -/53; F 6/55; m -/56; F 3/58; *F1 3/60; m -/56; M 1/63	1922	1930
425	*F1 7/56	1903	1917
426	*F1 10/59; *Fn 8/62; F 1/66	1931	1933
427	M 9/48; F 2/56; *F1 4/58; m -/59	1924	1928
428	*M1 9/58; M 10/60; M 1/63	1933	1933
429	M 1/59; *F1 12/60; F 12/63	1930	1930
430	F 3/53; *F1 4/56; F 1/65	1923	1922
431	*F1 4/58	1933	1933
432	M 1/60; *F1 5/62	1938	1937
433	*F1 6/56; F 6/57; M 4/59; F 11/62	1928	1927
434	*F1 6/56; F 10/58; M 2/61; M 5/66	1930	1930
435	*F1 10/61; F 8/63	1941	1941
436	M 5/53; *F1 12/54; Fsb 2/57; F 12/58; F 12/62; F 12/64	1933	1932
437	M 4/54; *F1 3/56	1923	1923
438	*F1 10/55	1923	1926
439	Msb -/33; F -/34; M -/36; F1 -/39; *F1 -/44	1908	1916
440	M1 5/58; M 8/60; *F1 9/62	1928	1928
441	F 5/49; M 1/52; *F1 3/53; M -/54; M -/55; F 8/59	1921	1925
442	M 5/58; *M1 1/60	1934	1935
443	Msb 12/53; *F1 12/54; F 7/56; *F1 10/57; Msb 1/59; F 7/62	1923	1928
444	*F1 3/64; M 4/65	1933	1937
445	m -/57; F -/59; M 12/60; *F1 8/62; F 12/64	1926	1927
446	Msb 3/61; F 7/62; *F1 8/63	1932	1936
447	*F1 12/55	1908	1912
448	*F1 9/64	1942	1941
449	*F1 1/55; F 1/57; M 6/63	1930	1930
450	M 5/61; F 4/63; *Fn 3/66	1934	1933
451	M 11/55; M 2/57; *M1 4/59; m -/60; F 12/63	1933	1933
452	*M1 10/59	1928	1927
453	F 3/65; *Fn 3/66	1945	1943
454	F 10/64; *F1 12/65	1936	1940
455	Fsb 11/52; *F1 2/54; M 7/57; m -/59; M 12/61; M 6/64	1929	1930
456	*F1 10/65	1938	1944
457	M 2/59; *F1 3/66	1922	1935
458	M 5/53; *M1 10/54; Msb -/58; Msb -/60; Fsb -/62	1925	1932
459	F11/58; *F1 7/62	1919	1930
460	*F1 5/60; F 8/64	1932	1933
461	*M1 6/64	1939	1939
462	*M1 8/59; *F1 5/62	1935	1939
463	*F1 4/61; m -/61; M 11/62; F 10/65	1931	1939
464	F 4/63; *Mn 2/66	1937	1943
465	*Fn 3/66	1944	1948
466	m -/53; F 11/54; M 11/56; m -/57; F 6/60; m -/57; *F1 9/64	1935	1934
467	*F1 9/61; M 3/63; M 8/65	1937	1938
468	Fsb -/51; M 3/53; m -/55; M 9/58; *M1 8/60	1921	1924
469	F 9/58; *M1 9/60	1931	1933
470	M 1/50; *F1 3/52	1920	1927
471	[F; F;] 1/56; F 7/57; *F1 12/60; M 5/62	1928	1932
472	M 8/36; M 6/38; M 8/39; F 3/42; F 11/43; F 4/45; F 11/46; M 10/48; *M1 11/49; M 4/55	1911	1916
473	m 8/45; M 7/48; M 10/50; *F1 10/57	1911	1914
474	*F1 1/42; F 12/45; m -/48; F 9/51	1912	1915
475	*F1 2/59; M 12/61; M 5/63	1930	1929
476	F 1/53; F 2/54; F 5/55; M 1/57; *F1 4/59; F 7/60	1924	1934
477	F 8/37; *F1 9/39	1911	1909
478	Fsb -/46; *M1 5/48; M 4/49; Fsb 6/53; Fsb -/54; F 8/57	1906	1914
479	[M; *M1] 9/41; M 9/47; F 11/51	1917	1918
480	*F1 1/65	1935	1941
481	*M1 8/44; [M; F;] 8/50	1920	1921
482	*F1 9/39	1920	1921

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Serial No.	Sibship	Date of Birth	
		Father	Mother
486	M 8/24; F 10/25; M 11/26; F 12/27; M 1/29; *F1 5/30; F 12/33	1899	1900
487	M 5/42; *F1 12/46	1914	1916
488	m -/31; F 4/32; m -/34; *F1 12/36	1895	1894
489	M 7/38; *F1 6/45	1911	1911
490	M 7/30; *M1 7/43	1900	1901
491	*F1 6/46; M 6/49; M 1/56; F 8/59	NK	1926
492	M 3/32; F -/33; M 5/35; *M1 6/41; m -/41; m -/-; m -/-; m -/-; m -/- (between 41 and 48)	1900	1904
493	M -/17; *F1 1/18; F 2/22	NK	NK
494	*M1 6/39	NK	NK
495	F 12/31; F 12/33; *F1 3/40	1899	1906
496	M -/18; *F1 6/20	1884	1884
497	*F1 9/38	1912	1913
498	*F1 3/41; F 8/43; m -/48	1903	1906
499	M 12/37; *F1 1/43; F 6/47; F 6/49; M 7/52; M 10/53; Msb 4/61	1914	1914
500	*F1 2/47; M 12/48; M 9/51; Fsb 4/53; Msb 7/55; M 10/60	1920	1926
501	m -/32; *F1 12/34; F 12/36; m -/38; F -/47	1913	1914
502	*M1 3/59; M 9/60	1930	1933
503	F 10/40; F 1/42; *F1 10/45	1905	1910
504	Msb -/-; F 2/40; *F1 6/46	1912	1920
505	M 1/40; F1 8/42; m -/46; F 11/47; *F1 9/49; F 3/54	1915	1917
506	*F1 8/38	1902	1907
507	*M1 2/45	1893	1906
508	*F1 12/47; F 5/51; F 7/55; M 1/60	1917	1924
509	M -/1897; M -/1899; F -/01; M -/03; F -/05; *F1 -/07; M -/09; F -/11	1869	1870
510	F 12/32; M 5/35; *F1 2/45	1905	1907
511	F 10/31; M 4/34; M 11/35; *F1 9/43; F 5/48	1903	1906
512	*F1 12/43; M 5/45	1907	1912
513	Fsb -/14; *F1 10/15; M 10/16; M -/18; F -/19; F -/20; M -/21; F -/22; M -/25; M -/27; M -/32	1890	1896
514	M -/1899; F -/01; M -/03; M -/05; M -/09; M -/-; *M1 6/12	1880	1900
515	M -/22; M -/28; *F1 8/39	1893	1893
516	F 1/35; *F1 6/36; F 1/38; F 1/42	1908	1912
517	M 7/39; F 1/41; *F1 10/42; M 5/44	1911	1913
518	*F1 8/42	1894	1910
519	F -/42; Msb -/-; *F1 5/47; F -/60	1922	1922
520	F -/22; M -/24; M 1/25; M 10/32; M 3/34; *F1 1/36	1897	1898
521	F 12/33; *F1 2/37	1902	1905
522	F 5/28; Msb -/33; *F1 4/36; F 6/38; F 12/44	1906	1906
523	*F1 10/43; F 1/45; [M; F:] 8/50	1912	1916
524	F 10/47; M -/48; *F1 9/49; M 8/53; M 7/55; F 11/61	1923	1922
525	*F1 2/47; M 8/48; F 9/49; M 3/53	1921	1922
526	F 5/37; M -/-; M -/40; *F1 3/42	1903	1914
527	M -/1897; M -/05; M -/06; M -/08; M -/10; *F1 1/12	NK	NK
528	M 5/43; *F1 10/46; M 11/49	1919	1922
529	*M1 11/28	NK	1909
530	*F1 7/33	1898	1901
531	F 11/37; F 2/40; *F1 8/43	1908	1909
532	*F1 6/48; M 4/53; F 5/59	1920	1925
533	*F1 -/33	1906	1907
534	*F1 11/46	1910	1913
535	F 8/31; M 10/32; *F1 3/37; F 10/41	1902	1908
536	M 7/47; *F1 8/41	1915	1912
537	M -/-; *F1 9/40; M -/-; [M; M] -/-	NK	NK
538	F 3/36; M 3/39; M 10/42; *F1 3/44	1908	1915
539	*F1 4/49; M 7/51; M 6/53; M 4/55; M 4/59; F1 4/61; M 11/62; [F; F] 3/65	1917	1923
540	F 9/30; *F1 12/31; M 5/32; F 7/35; F 8/37; M 12/41; F -/43; m -/48	1906	1913
541	*F1 3/47; *F1 2/49; F 4/51; M 7/54; M 7/55	1917	1922
542	M 7/21; F 6/24; *F1 9/34; m -/36	NK	1894
543	*F1 8/30; M 3/32	1898	1902
544	*F1 6/27; F -/29; F -/33; F -/36; M -/38	1902	1904
545	M 2/46; M 3/47; M 5/49; *F1 6/50; M 9/52; F 3/54	1923	1923
546	F 7/17; F 12/19; M 11/24; *F1 7/27	1888	1889
547	*M1 10/36	1908	1912
548	M1 8/20; F -/22; M -/24; M 4/26; [M; F; *F1] 4/29	NK	NK
549	M 3/43; F 1/47; F 5/48; M 7/49; *F1 10/50; M 10/52; Fsb -/-	1923	1923

Appendix—continued

Serial No.	Sibship	Date of Birth	
		Father	Mother
569	*F1 2/37; F 4/42; M 7/45	1910	1915
570	F 1/36; *F1 1/40	1913	1914
571	*F1 2/34; F 10/35; F 12/36; M 6/39; M 8/43	1913	1915
572	M 9/35; F 12/36; *F1 8/38	1928	1916
573	*F1 12/47	1917	1918
574	*F1 3/49; F 2/53; F 2/54; F 1/57; Fsb 3/64	1921	1922
575	*F1 8/37; m -/38	1904	1911
576	F 5/45; F 6/46; *M1 10/47; M 6/50; F 5/61; F 12/63	1922	1923
577	M 6/34; M 8/38; *F1 12/40; m -/42	1899	1904
578	F 6/38; M 12/42; *F1 3/46	1909	1910
579	M 8/26; F 2/27; F 3/28; F 5/29; M 10/31; M 10/32; F 7/34; F 2/35; F 7/36; F 1/38; *F1 3/39; F 7/41; F 1/43; F 11/48	1899	1903
580	*F1 7/42	1895	1905
581	M -/25; M 1/27; *F1 1/31; M 5/33	1898	1900
582	F 9/23; M 3/25; M 5/26; F 1/29; F 8/31; *F1 10/33	1892	1895
583	M 10/30; F 11/33; F 7/41; *F1 11/47; M 12/51	1906	1908
584	*M1 2/50; F 12/55	1926	1926
585	F 3/17; *F1 10/23	1890	1891
586	*F1 12/31	1898	1901
587	F -/08; M -/09; F -/11; F -/12; *F1 11/15; M -/17; F -/19; M -/21	1880	1883
588	*F1 7/35	1888	1890
589	*F1 5/21; M 6/24	1895	1896

Serial No.	Children	Date of Birth	
		Father	Mother
214	m -/63; m 1/64; Fn 12/65		*1938
216	M 10/55; F 4/58		*1925
227	F 10/56; M 2/58		*1930
229	M 8/42		*1915
230	F 2/48; F 12/58		*1916
245	M 4/66		*1942
248	M -/47; *F1 9/48; M 6/51 (See 249)	1927	*1926
249	F 8/66		*1948
260	Fn 8/64		*1940
272	F -/28; M 7/30; M 6/34		*1902
283	M 12/54; M 9/57		*1927
284	M 12/60; F 10/65		*1937
293	F 3/61; m -/61; m -/62; m 10/62; m 1/64		*1941
297	*Fn 8/59; M 9/60; F 8/63; *Fn 3/65 (see 64/65)	1936	*1935
383	M 3/64		*1935
477	F 9/66		*1942
484	M 5/67	*1944	
485	Mn 10/62; F 3/67	1937	*1939
486	M 7/67; F 7/66		*1930
488	M 4/60; F 6/63		*1936
491	M -/65		*1946
493	M 11/46		*1918
495	M 8/66		*1940
497	Fsb 8/62; M 2/64		*1938
499	M 1/63; F 4/64; M 2/68		*1943
501	F 2/57; F 4/58; M 8/61		*1934
506	Fn 10/64		*1938
511	m -/66		*1943
512	M 7/68		*1943
513	M -/43; F -/42		*1915
514	M 10/38; M 10/41; F 11/44	*1912	
515	Mn 8/58; Fn 4/60; F 7/64		*1939
516	M 3/61; M 3/65		*1936
517	M 10/65		*1942
520	M 8/60; F 4/63		*1936
521	F 2/61; m -/63; m -/64; Fn 9/66		*1937
522	M 3/58; M 10/59		*1936
523	Fn -/—		*1943
526	F 5/63; M 6/64		*1942
527	F 7/31; m -/31; F 2/33; F 7/37		*1912
530	F 9/58; F 3/64		*1933
533	M 12/66		*1933
536	F 5/65		*1941

Appendix—continued

Serial No.	Children	Date of Birth	
		Father	Mother
537	M 5/63; F 7/65		*1940
538	M 11/66; F 10/67		*1944
540	F 10/54		*1931
541	M 7/66		*1947
543	F 10/62; F 12/67		*1934
544	F 11/56; M 5/58; M 12/60		*1930
545	F 15/61		*1927
547	m 9/67		*1927
549	F 11/56; F 11/58		*1929
555	F 6/62; M 8/64		*1937
556	M 10/57; M 4/62		*1931
557	F 3/59; F 7/60; F 5/64		
560	M 10/54; M 11/56; F 8/64	*1927	*1927
561	F 5/66		*1943
562	M 7/56; F 10/58; M 2/62; F 10/63; m 1/67		*1925
563	m -/-; m -/-		*1942
566	F 5/63; M 7/64; M 10/65; M 12/66		*1943
567	F 1/66; F 11/67		*1941
571	M 10/61		*1934
572	F 5/61; m -/62; m -/63; F 1/65		*1938
575	M -/60; F -/62		*1937
577	M 5/64; M 7/65; F 2/67		*1940
579	F 8/62; F 2/64		*1939
580	M 10/67		*1942
582	F 1/64; F 11/65		*1933
585	F 6/58		*1923
587	F 4/41; M 9/42		*1915
589	F 11/45; F 4/51		*1921