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Birth Order Studies: Some Sources of Bias

By J. S. PRICE and E. H. HARE

"... they both felt, after the conclusion of their work, very doubtful as to the possibility of definitely proving the existence of a real differential incidence of any character in order of birth. The whole question seemed so open to fallacious possibility in different directions."

[Report of comment by G. Udney Yule in the discussion following his joint paper with M. Greenwood at the Royal Statistical Society in 1914.]

Recently there has been an upsurge of interest in the association between behaviour, both normal and abnormal, and birth order. For instance, Barry (1967) has pointed out that in certain societies excessive parental demands are made on the first-born child, and "excessive parental demands could lead to superior achievement in a robust individual or to psychiatric illness in a child who was especially vulnerable . . ."; thus putting into modern language an interesting genotype–environment interaction which was summed up in the ancient Chinese proverb "fire burns wood but tempers iron".

Barry's hypothesis is based on the finding of an excess of schizophrenia in the first-born, not only in some oriental countries but among upper class Americans. In lower class American families, on the other hand, the excess appears to be in the later-born positions. Of course, social class is related to family size, and from this point of view the schizophrenics show an excess of first-born in small sibships and of last-born in large sibships. These, and other findings relating to birth order in psychiatric patients, may be due to differences in susceptibility to mental illness of individuals in the various birth ranks, or they may be due to one or more of several biases which are liable to affect the data.

The history of birth order studies is not a happy one. The subject matter is particularly difficult to think about clearly, even for the mathematician. There has been much controversy about the methods which can be used to determine whether the observed distribution of birth ranks in a sample differs from the expected distribution. In 1914, for example, Greenwood and Yule pointed out a serious fallacy in the method used by Pearson and his colleagues (e.g. Heron, 1907); and over 50 years later attention has been drawn to a fallacy in the method suggested by Greenwood and Yule (Barker and Record, 1967*a*). In the present paper we will not consider this aspect of methodology, but rather various biases which may distort the observed distribution of patients by birth-rank. These fall under four main headings. Firstly, there are biases in the sample due to biases in the population from which the sample is drawn. Secondly, there is the distortion introduced when sibships are not complete. Then there are the biases which the investigator may introduce in his manipulation of the data. And finally, there is the possibility of a correlation between birth order and the proportion of missing or unreliable information. Most of these sources of bias have been pointed out before and have been reviewed by several authors, such as Greenwood and Yule (1914), Gregory (1958), Chen and Cobb (1960), and Barker and Record (1967*b*). However, the pitfalls of the subject are so great that a further and perhaps more systematic review of this particular aspect of birth order studies seems justified.

Complete Sibships

Let us first of all consider a sample of patients whose sibships are complete; that is, a sample whose mothers have all passed the end of the reproductive period. For convenience we will call all people from complete sibships "adults". Let us also assume that these adult patients are suffering from a disease which obeys the "null hypothesis"; that is, that there is no correlation between the disease and birth rank; or, in other words, that people of all birth ranks are equally likely to develop the disease *and* to be included in a sample of patients suffering from the disease. In such a sample, with certain qualifications, we expect the patients in each size of sibship to be randomly distributed between the various birth ranks (first, second, third, etc.); and over the sample as a whole we expect there to be approximately as many first-born as last-born, and as many early-born (from the first half of the sibship) as later-born (from the last half of the sibship). This is obvious if one considers the whole population, because for every sibship of two there must be one first-born and one second-born, and so on. The sample of patients will reflect the random distribution in the general population, provided that the character of being a patient is not correlated with birth rank.

It has been supposed, mistakenly, that since the chances of developing a disease increase with age, a sample of patients will contain more older (early-born) siblings; but a little reflection will show that this is not so, provided that the inclusion of a patient in the sample does not lead to the inclusion of affected siblings in the sample too (Gregory, 1958). If two or more patients from the same sibship are independently ascertained, they may be included in the sample without introducing any bias. But if, as might happen with a rare disease in which every effort is being made to increase the number of cases, some patients are included in the sample as a result of being siblings of patients who have already been ascertained, then, if the patients are being studied before the risk period of the disease is passed, the inclusion of such extra siblings will result in an excess of older (early-born) patients in the sample; because, of course, the early-born siblings will have passed through more of the risk period than the later-born siblings, and therefore more of them will have manifested the disease and more extra early-born than later-born siblings will be added to the sample.

In certain circumstances, the principle of random distribution of adults by birth rank in the general population may not hold good. There are two main causes for this: a change in the reproductive habits of the population, and a change in the birth rank distribution of the population between birth and the age range from which the sample is selected. Both these points require some elaboration.

1. *Changes in the Number of Births from Year to Year*

J. A. Cobb (1914) pointed out that if a man is born in 1870, say, and has, or had, a brother five years older or younger, then as there were more births in 1875 than in 1865 the brother is more likely than not to have been born in 1875 and therefore to be the younger of the pair. To take it to an extreme, if there were no births at all during the 30 years preceding 1870, then the brother must be younger and the man himself must be first-born. As a general rule, then, we may say that when the number of births is increasing the population will contain an excess of first-born and other early-born (from the first half of their sibships); and conversely, when the number of births is falling the population will contain an excess of last-born and other later-born.

The number of births may change either because of a change in the number of families being started or because of a change in the size of families. Both kinds of change have been taking place in our population over the past 80 years and it may be helpful to consider their effects separately. The first alters the ratio of early-born to later-born in the whole of the sample. The second also has an effect on the whole sample, but its main effect is seen when the sample is subdivided into small and large sibships, since it has an opposite effect on the two subdivisions. It will be seen later that a decline in family size produces much the same effect as incompleteness of the sibships.

The number of families started is closely related to the number of marriages. In Scotland in 1945, for example, 34 per cent. of first-born children were born within one year of marriage, 63 per cent. within two and 75 per cent. within three years (Registrar General for Scotland, 1945). It can be seen from Fig. 1 that marriages in England and Wales have increased steadily from 1881 to 1950, since when they have levelled off. It is possible to calculate roughly the effect of this rise on the relative number of first-born and last-born in a sample

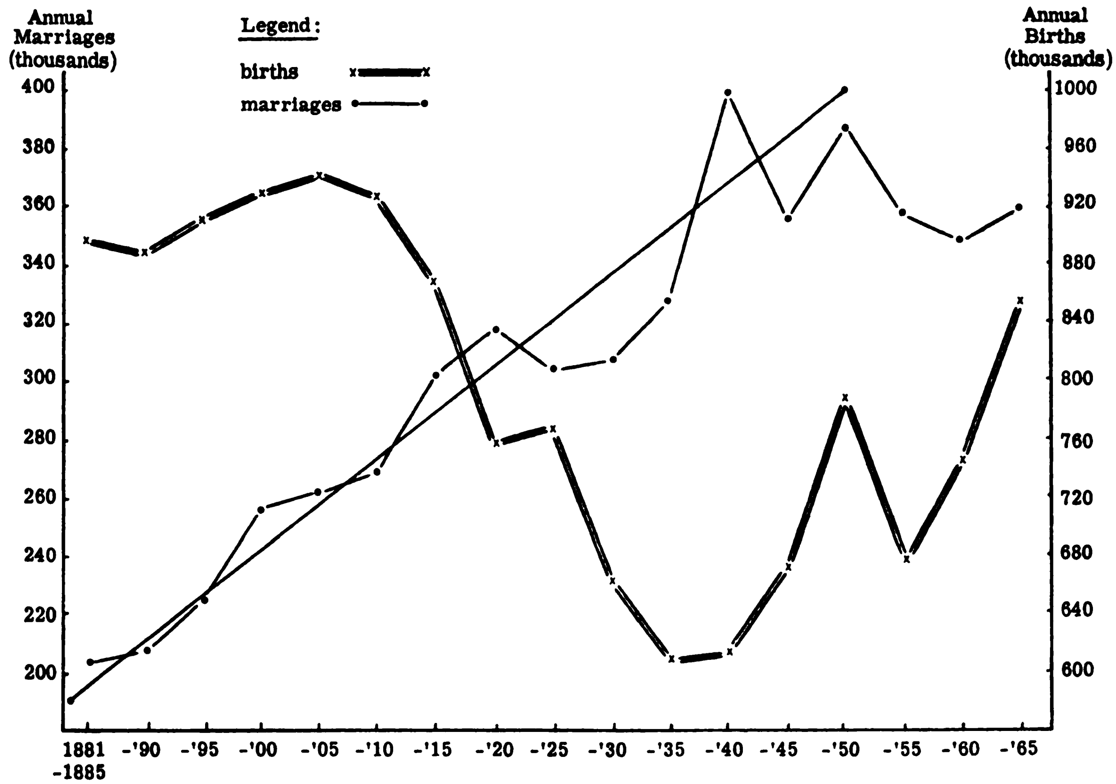

FIG. 1.—Marriages and births in England and Wales for five-year periods 1881–1965 (Registrar General, 1954; 1967). The straight line is discussed in the text.

born during this period. Let us assume for simplicity that the span of all sibships, from first-born to last-born, is 10 years, and that a first-born arrives two years after every marriage. If in 1967 we consider all the people in England and Wales aged 50, we will find the following number of first-born and last-born (assuming none have died, etc.):

First-born. In 1915 there were 290,000 marriages (using the straight line in Fig. 1, drawn by eye, to even out the fluctuations) and therefore according to our convention there were 290,000 first-born born in 1917, who will be aged 50 in 1967.

Last-born. In 1905 there were 258,000 marriages, and therefore, with a sibship span of 10 years, there were 258,000 last-born in 1917, who will be aged 50 in 1967.

The ratio of first-born to last-born in the general population is thus 290,000 : 258,000 or 1·124 : 1. The sample of patients aged 50 can be expected to exhibit the same ratio. The ratio will vary a little with age, since the increase in marriages is linear and not compound, but a sample of psychiatric patients over 15 will have a ratio of about the same order. In a sample of 4,000 patients, assuming half to be either first- or last-born, such a difference would be significant at the 1 per cent. level. (It is interesting to note the relatively large number needed to detect a 12 per cent. excess of first-born; if the general population distribution were unbiased, and first-born were 12 per cent. more likely to become psychiatric patients than last-born, it would require, on average, a similar sample of 4,000 to demonstrate the difference at the 1 per cent. level of significance.)

A similar though less marked effect occurs with the intermediate birth-ranks, a rise in marriages resulting in an excess of early-born over later-born.

From 1938 onwards the Registrar General has been presenting the annual births subdivided by birth-rank (in Part 2 of the Statistical Review), so that in the future it will not be necessary to infer the number of families started from the number of marriages. Whichever source of data is used, it would be as well to take it as a rather general guide, unless the sample being studied matches the general population very closely in social and ethnic variables. If matching were good, or if one were able to ascertain all cases of a disease in England and Wales, the birth ranks of the patients could then be compared directly with the birth ranks of the general population for the appropriate years of birth. This possibility will not be explored further, as it is unlikely to be practicable in the case of psychiatric illness.

During most of this time when the number of marriages has been rising, the number of births has been falling (Fig. 1), and this is due to the *reduction in family size* which has occurred during the same period.

"The period up to the Second World War saw 'marriage cohorts' (i.e. women married in a particular year) with steadily decreasing family sizes. These ultimate family sizes decreased from 6·16 for women married in 1861 to 1869 to 3·30 for those married in 1900 to 1909 and to 2·08 for those married in 1929" (Registrar General, 1965; see also Table VII in Gregory, 1958). Family size reached its nadir in 1940, and has been rising steadily since then.

It is easiest to comprehend the effect of declining family size by considering a highly simplified population. Let us imagine a population in which 100 families are started every year. Ten per cent. of the mothers have red hair, and all these red-haired women have 20 children at one-year intervals. All the other women have 10 children each at one-year intervals. The breeding pattern is stable so that 100 families are completed every year. Now let us indulge in the fantasy that in 1920 a red-haired primipara's club is started to which all red-haired women having their first child in 1920 or subsequent years belong. Family planning is in vogue and a group norm is immediately established that members have exactly five children at one-year intervals. All members are strict conformers to the group norm.

We can examine the resulting population with the advantage of hindsight. In 1919 the red-haired women produced 10 first-born and 10 last-born (in sibships of 20) and the others produced 90 first-born and 90 last-born (in sibships of 10). In each of the years 1920 to 1923 inclusive, the red-haired women produced 10 first-born (in sibships of five) and 10 last-born (in sibships of 20). The other women continue as before to produce 90 first-born and 90 last-born in sibships of 10. In the population born during these years there is no disturbance in the overall ratio of first-born to last-born, but there is a marked excess of first-born in small sibships and last-born in large sibships. During each of the years 1924–1938 inclusive, the red-haired women produced 10 first-born (in sibships of five) and 20 last-born (10 in sibships of five, and 10 in sibships of 20). During these years there is thus an overall excess of last-born, a marked excess of last-born in large sibships but no difference in small sibships of five. Taking the whole population born 1919 to 1938 we find the following:

	First-born	Last-born	Ratio of First-born to Last-born
Sibships of 5	190	150	1·27
Sibships of 10	1,800	1,800	1·00
Sibships of 20	10	200	0·05
All sibships	2,000	2,150	0·93

From this highly simplified and exaggerated model, the overall effect of a reduction in family size can be seen:

(*a*) A slight overall excess of last-born.

(*b*) A moderate excess of first-born in small sibships.

(*c*) A marked excess of last-born in large sibships.

The same effects can be seen from the model in Table III. This model shows the effect of studying a sibship before the last sibling has been born; but if the last sibling is not born at all the result is of course the same. The two separate phases (outlined above) can be observed, one in the cohort enclosed in the rectangle and the other in the cohort following.

Family size is now increasing in England and Wales, and the effect of this will be the reverse of the above. Evidently if a sample of patients is drawn from a wide age range, so that their years of birth include both increasing and decreasing family sizes, the two biases will tend to cancel each other out.

2. *A Correlation between Birth Rank and Movement into (or out of) the Population*

The two factors we will consider here are migration and death.

Most birth order studies are carried out in large cities or university towns, and a relatively high proportion of the subjects will be immigrants from abroad or at least from other parts of the same country. Perhaps the days are past when the eldest son stayed to till the family plot of land while the younger sons came to the city to seek their fortunes, but it might be premature to assume that population movements are independent of birth rank. Younger children, for instance, lose their parents at an earlier age; and it has been suggested (Hill and Price, 1967) that the loosened family ties of such bereaved children may facilitate migration. Three control groups studied in London by Dennehy (1966) showed an excess of parental bereavement in childhood, possibly reflecting an excess of later-born siblings. Fortunately, it is easy to control for this bias: if birth-place is recorded during the collection of the data, a separate analysis can be made for natives and immigrants.

Another change in the population which may be related to birth order is death in childhood. Newcombe (1965) studied the relation between death in infancy and pregnancy order, using the British Columbia record linkage system. Compared with all live births, child deaths increased with increasing pregnancy order; the effect was greatest from one month to one year and for pregnancies after the fifth. The births of subsequent siblings could not be taken into account, and therefore it is not known whether the trend would hold good if sibship size were held constant. It would be reasonable to suppose that it would. Deaths from rhesus incompatibility, for instance, increase with birth-rank for any sibship size, and maternal care is likely to deteriorate towards the end of a sibship. Schreider (1967) has pointed out a further possible source of disadvantage for later-born children; he studied the rise in serum iron of 32 pregnant women, and showed a correlation of $-0 \cdot 55$ between serum iron and number of previous pregnancies. For a number of reasons, therefore, it seems likely that more later-born children die in childhood, and therefore, the surviving population will contain an excess of early-born.

The differential death rate falls rapidly after the first year of life, so that most of this bias could be excluded by counting as a sibling only those individuals who have survived to the age of one. This, of course, excludes from the analysis any living siblings under the age of one. If any of the patients in the sample do in fact have siblings under one, the sibships are not likely to be complete, and excluding those under one will have the effect of making them even less complete. The biases outlined in the next section will, therefore, be increased. In such a case the choice of procedure will depend on a weighing-up of the relative importance of the two sources of bias.

Incomplete Sibships

When one examines the birth order of a population of children or young adults, it is likely that some members will come from incomplete sibships; that is, their mothers will have further children after the date of the investigation. This fact disturbs the random distribution of individuals by birth rank. Let us consider, first of all, what sort of biases are produced and how they arise, and then how their importance in any given population may be assessed.

We are fortunate in having data from an actual population to present as an example. Table I is derived from the report of the Scottish Council for Research in Education (1949), and gives birth-rank by sibship size for practically all the children attending grant-aided schools in Scotland who were 11 in June, 1947.

TABLE I

Birth-rank and sibship size of Scottish children aged 11 in June 1947. The total population of 11-year-old children in Scotland at this time was estimated by the Registrar General as 80,300. (Reference in text.)

Birth rank	Sibship size 1	2	3	4	5	6	7	8	9	10	Total
1 ..	8,239	9,768	5,779	2,766	1,181	454	163	53	17	4	28,424
2 ..		6,998	5,340	3,027	1,672	782	316	102	36	5	18,278
3 ..			4,319	3,055	1,781	1,022	556	204	72	25	11,034
4 ..				2,730	1,822	1,049	624	356	121	48	6,750
5 ..					1,729	1,028	643	407	241	106	4,154
6 ..						1,068	636	423	234	113	2,474
7 ..							660	362	221	119	1,362
8 ..								377	208	138	723
9 ..									226	97	323
10 ..										108	108
Total ..	8,239	16,766	15,438	11,578	8,185	5,403	3,598	2,284	1,376	763	73,630
									Greater than 10		756
									Not known		825
											75,211

This material has already been discussed from the point of view of birth order by Berg *et al.* (1967). We can expect many children of 11 to come from incomplete sibships, and it is very evident from Table I that the distributions are far from random. Each sibship size is represented by a column, and when sibships are complete we should expect all the cells in a column to be approximately equal. But in sibships of two and three we have a marked excess of early-born, and in sibships of six and over we have an even more marked excess of later-born. There is an overall excess of first-born over last-born; this is an unexpected finding, and will be discussed in a later section.

If one looks at Table I, it is clear that the children in some cells of the matrix are more likely to come from incomplete sibships than others. For instance, 108 children are tenth in sibships of 10. This means that 11 years ago their mothers had a tenth child, since when they have had no more children. It is almost certain that these mothers are past the childbearing age and that these sibships are complete. The case is quite different with the four children who are first-born in sibships of 10. These mothers are clearly very fertile and probably started their families at an early age. Now they are likely to be in their early thirties or even late twenties, and probably had their last child only a year or two ago. There is every likelihood, therefore, that they will have one or even several more children, and these four first-born will end up as first-born in sibships of 11 or more.

As a general rule, then, the earlier the birth rank a child is in, the more likely is the mother to have more children and thus for the sibships to be incomplete. Another important factor is the time that has elapsed since the mother had her last child. For a child in any given birth-rank, the chances of the sibship being complete are greater when the child is towards the end of the sibship; if he is last, his mother has not given birth for 11 years, and after this length of childlessness the chances of further children are much reduced.

This correlation between "completeness" of sibship and the time since the last member of the sibship was born will be of importance in considering the reason for the excess of early-born in small sibships. Take the first-born in sibships of one and two, for instance. The mothers of these children all had their first child 11 years ago; one group has had a further child, the other has remained without further children for 11 years; which will have more children in the future? We would expect, on general grounds, that the mothers who have been longer childless will have less children, being probably older than the others; but we cannot be certain, because the two probabilities are to some extent independent. If, for example, it was considered shameful in the population to have only one child, and downright disgusting to have more than two, then the mothers of the only children would probably be our best bet for further births. Fortunately, the Registrar General for England and Wales provides empirical data on the number of births per year for cohorts of women married in certain specified years, subdivided by the number of children they have already had (Part 3 of the Annual Statistical Report). These Tables show that child-bearing goes on longer the more children the mother has already had, roughly at the rate of two years per child. Although these mothers are matched for year of marriage rather than year of first child, the two events are so closely related that it seems reasonable to conclude that, according to our expectation, for any birth rank the chances of incompleteness of the sibship will increase with the size of the sibship.

Let us now consider the biases in more detail.

Overall Excess of First-born

Shortly after they were born, all these Scottish children were of course last-born (ignoring twins). If we had drawn up Table I at that time, they would all have appeared in their present rows but would have been crowded into the left hand cell of each row; that is, they would all have been in the principal diagonal, stretching from only children to tenth-born in sibships of 10, which contains all the last-born. As further children were born in their sibships, they would have migrated towards the right along the cells of each row, and we would expect this process to go on until there were just about as many last-born in the lower diagonal as first-born in the top row. At no stage should the number of last-born fall below the number of first-born.

However, in the case of the Scottish school-children, there are 28,426 first-born and only 26,548 last-born, giving a first-born–last-born ratio of 1·071. This difference is large and highly significant, too large to be explained by the 5,000-odd children who were living in Scotland but not included in the sample (presumably because they were not attending grant-aided schools) even if it were thought that such exclusions would contain an excess of last-born, which on the face of it seems unlikely.

We expect an excess of last-born but in fact find an excess of first-born. How are we to explain this paradox? Clearly it must be due to one of the biases discussed in the previous section. It is too great to be accounted for by the increased childhood mortality of last-born. With children of this age, it is not likely to be due to last-born going off to America to seek their fortunes. It is not due to a change in family size; in the 'thirties family size was still falling, and this would tend to give an excess of last-born rather than first-born. It must therefore be due to the increase in families being started, as reflected in an increase in marriages. We have seen earlier from the English data that the rise in marriages would give a first-born–last-born ratio of 1·124, which is enough to cover the observed ratio of 1·071 in the Scottish children and allow a little for incomplete sibships. In fact, we probably do not need to allow much for an excess of last-born due to incomplete sibships; as we have seen earlier, the incomplete sibships in this population involve mainly the early-born, and not many mothers will have a further child after a lapse of 11 years. This is just as well for our argument, because Scottish marriages have not increased as much as English marriages and there was a marked increase after the First World War which would have affected this population a little. Nevertheless, the majority of first-born in the Scottish cohort were probably born of parents married in 1934 and 1935, and from the data in Table II it can be seen that marriages in Scotland were higher then than for any period in the preceding 13 years.

TABLE II

Marriages in Scotland in the years preceding the birth of the 1936 cohort. (*Registrar General for Scotland, 1968.*)

Years	Marriages
1921–25	34,720 (Annual average)
1926–30	32,605 (Annual average)
1931	32,652
1932	33,157
1933	34,201
1934	36,934
1935	37,997

Incidentally, this excess of first-born not only conflicts with the bias expected from incomplete sibships, but also flouts Cobb's law. Earlier we mentioned Cobb's deduction that a rising number of births is associated with an excess of first-born, and a falling number of births with an excess of last-born. But in the case of the Scottish children, the excess of first-born occurred against a background of falling number of births. In the decade 1926–1935 there were 929,900 births in Scotland compared with 912,140 births in 1937–1946 (Registrar General for Scotland, 1968). In this case an increase in marriages and a reduction in family size are acting in opposite directions on the first-born–last-born ratio. But the change in family size, as we have seen earlier, has only a slight effect on the ratio, and therefore it is not sufficient to balance the effect of the increase in marriages, even though it outweighs the latter when it comes to the total number of births.

Excess of Later-born in Large Sibships

This is the largest bias and the one which is likely to be present to a significant extent in a population of adolescents or young adults. We have seen earlier that whereas the later-born children (lower rows of Table I) are likely to come from complete sibships, the early-born have younger mothers and their sibships are more likely to be incomplete. Thus there is likely to be a migration towards the right in the upper rows of the table, so that the excess of later-born in the large sibships will become balanced by those who are now early-born in small sibships. As Yule pointed out after the discussion of his paper with Greenwood in 1914, it requires some time to become the first-born of a sibship of 10, and it is not surprising that not many have achieved it by the age of 11.

A similar bias is given by a reduction in family size, and part of the excess of later-born in the large sibships of Table I is probably due to this.

Excess of Early-born in Small Sibships

This bias seems rather unexpected at first sight, and its presence is contingent on the rates at which women of different parity have children. But we have seen above that the mothers of the first-born in sibships of two, having had their first child 11 years ago and one subsequent child, continue to have children later than either the mothers of only children or the mothers of the second-born of sibships of two, both of which groups of mothers have been childless for 11 years. There will be a time, therefore, when some of the first-born in sibships of two are due to become first-born in sibships of three, whereas the second-born in sibships of two and the "only children" are all from complete sibships. Since in the final state there will be equal numbers of first-born and last-born in sibships of two, there must be an excess of first-born at least for the last year or two before the final first-born become first-born in sibships of three.

This rather complex argument can, perhaps, best be appreciated by the model breeding population illustrated in Table III, which is based on those used by Barker and Record (1967*b*). In this highly artificial population with constant birth interval and only one final sibship size, there is, at the age of 11, a clear excess of early-born in the smaller of the two sibship sizes which then exist. The same applies if larger families or mixed family sizes are considered.

This distortion of small incomplete sibships may be of some importance. There is a temptation to consider only a single sibship size in studying birth order, e.g. sibships of three (Grosz and Miller, 1958), because of the great simplification of the calculations. If sibships are incomplete, a false excess of first-born may

TABLE III

*A model population of 3-child families breeding at 7-year intervals: each family is depicted on a separate line, and the numbers refer to the birth-rank. The children, who are 11 years old at the time of the investigation, are enclosed in the rectangle. There are two children from 3-child families, (marked *) so that $\frac{2}{3}$ child would be expected in each birth rank; in fact there is none in the first birth-rank and one in each of the second and third. There is one child from a 2-child family (marked †) [the third child in this family had not been born at the time of the investigation], so that we would expect $\frac{1}{2}$ child in each of the two birth-ranks. In fact the child is in the first birth-rank. Thus a sample of such cohorts would contain more first-born children than expected in sibships of two (small sibships) and more last-born than expected in sibships of three (large sibships, relatively).*

			Cohort	Date of Investigation		
1	2	3				
	1	2	3*			
		1	2*	3		
			1†	2	3	
				1	2	3

Hypothetical 7-year birth intervals.

appear, or a real excess of last-born be obscured. To take an extreme case, we might take a sample of children who pass the 11-plus and study birth order separately for small and large sibship sizes. If, as is likely, we obtained a distribution similar to that in Table I, we might be tempted to conclude that in small families the first-born are at an advantage (private coaching from parents) while in large families the last-born are favoured (private coaching from older siblings).

Conclusions about Incomplete Sibships

When sibships are incomplete, there is an excess of early-born in small sibships, an excess of later-born in large sibships, and an overall excess of later-born. These effects are the same as those produced by declining family size; in fact, whether the tail-ends of some sibships are cut off by family planning or by the fact that the population is studied before they have been born makes little difference to the statistics. The important point is that the two sources of bias are both likely to be operating in a population of young adults studied today, and they will thus augment each other. In a few years, when patients have been born at a time of increasing family size, they will tend to cancel each other out.

At what age does the completeness of sibships cease to become a problem? A woman's capacity for childbearing does not last for more than about 30 years, so that, in patients over 30, sibships will certainly be complete (a sibship is usually defined as all the children born alive to the patient's mother). From figures published annually by the Registrar General (Part 3 of the Statistical Report) it is clear that after 25 years of marriage the fertility of women (married once only) is negligible. At the age of 11, as we saw with the Scottish children, the distortion is very marked. If one were to investigate birth order in a population under 15, say, some control for incomplete sibships would be essential. In the case of patients between 15 and 25 the position is more doubtful, particularly if only a proportion of the patients fall into this age range. Here one could adopt the strategy of Granville-Grossman (1966*b*) and analyse the over-thirties separately from the under-thirties. If, on the other hand, it were thought that birth order effects might be more important in the younger group, it might be wise to adopt one of the tactics described later.

Some idea of the incompleteness of the sibships may be obtained from data on sibship span (the interval between the birth of the first and last member of the sibship). For demographic reasons the investigator would be wise to use his own population to calculate sibship span, but some of our own data are shown in Table IV. Patients were asked the ages of the first and last children born to their mothers, span being the difference between these ages.

7·4 per cent. of sibships span more than 20 years, and therefore of first-born aged 20 we can expect 7·4 per cent. to come from incomplete sibships. Allowing a birth interval of two years between first-born and second-born, we can also say that 7·4 per cent. of second-born aged 18 will come from incomplete sibships, and so on for each birth rank and age group in the period of risk. These figures are probably underestimates, because some of the

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TABLE IV

Age span of the sibships of 363 patients. The patients are consecutive adult admissions to the Maudsley and Bethlem Royal Hospitals, September, 1967, to March, 1968, age range 16–65. Excluded from the data are 49 patients who were only children and 21 who were not able to give the ages of their oldest and youngest siblings.

Years	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 & over
No. of Patients	29	58	45	42	43	29	26	32	18	14	13	9	5
Cumulative %	8·0	24·0	36·4	47·9	59·8	67·8	74·9	83·5	88·7	92·6	96·1	98·6	100·0

sibships in Table IV must be incomplete, and long spans are probably over-represented in the "not knowns". Precision could be increased by calculating span of sibship separately for each size of sibship (the present data are too few for this) and also by taking other variables into account, such as the age of the mother and the age of the youngest member of the sibship. The proportion of incomplete sibships can then be assessed in relation to the object of the study and the magnitude of the birth order effect which it is desired to detect.

EXCLUSIONS OF SIBLINGS OR SIBSHIPS

Sibships may be incomplete in another way. Although all the siblings may have been born, some may not have lived through the whole period of risk of the disease being considered. This becomes important if the investigator wishes to exclude some of the siblings or whole sibships from the data.

Thus, Dalen (1965) argued that familial cases could with advantage be excluded from a birth order study, on the ground that these cases are likely to be genetically determined and so their presence might obscure some environmental influence correlated with birth order in the non-familial cases. Or again, it might be thought reasonable to exclude a sibship of two in which both members are affected, on the ground that such a sibship provides no information about the relation between birth order and the disease.

Yet such exclusions would introduce a bias. For, since the chances of having manifested the disease increase with age, more older siblings than younger siblings will have contracted the disease, and therefore more later-born than early-born patients will be excluded There will thus be an apparent excess of first-born patients and patients from the first half of their sibships. The parents, uncles, etc., of later-born patients are all on average older than those of early-born patients, and thus more likely to have manifested the illness, so the exclusion of any "familial cases" will lead to a similar false excess of early-born patients. The effect is similar if ill siblings are excluded from the sibships; for when older siblings are excluded, the patient appears more "early-born" than he really is and there will be an apparent deficit of later-born patients. The same argument applies to the exclusion of siblings who have died, since death is also correlated with age, and is more likely to have affected the older than the younger siblings. If, however, the whole study could be carried out retrospectively on patients admitted many years ago, whose siblings had all died, it would be reasonable to exclude siblings who had also had the disease and who had died before a certain age, since the chances of having done either would be independent of birth order. Such a study would of course be impracticable in this country because of the difficulty of following all the siblings to the end of the risk period.

These arguments apply not only to studies of birth order and ordinal position, but also to studies of variables which are correlated with birth order, such as parental age and bereavement, in which the siblings are used as controls. For instance, Granville-Grossman (1966*a*) in a study of parental age in schizophrenia, excluded siblings who had either died or developed schizophrenia. He also excluded siblings under the age of 16, which introduces the same bias as is found with incomplete sibships.

Exclusion of siblings is, of course, acceptable if the criterion of exclusion is as likely to exclude a younger as an older sibling. This is the case with the ingenious method described by Barker and Record (1967*b*) to control for incomplete sibships, changes in population size and other complicating factors. The method is designed for the study of congenital anomalies and other diseases in which complete ascertainment in a population is possible, and some modifications would be necessary if it were to be used for adult psychiatric patients. The method requires a knowledge of the ages of all siblings.

This caveat about excluding patients or siblings also applies to any other manipulation of the data which is liable to upset the random distribution of birth ranks. It has been pointed out that the sexes may not be uniformly distributed by birth rank (Loxton, 1962), so that a breakdown by sex may introduce an artefact into the results.

Exclusions Because of Incomplete Data

In the first study of birth order in mental illness, published by the Eugenics Laboratory in 1907, Heron wrote:

> "What appears from the statistical side at present so urgent is the need that those who have not only the opportunity but the clinical training necessary for accurate observation should record their facts in a form in which the trained statistician can apply to them the methods of modern statistics."

This advice, along with his strong plea for a central register of psychiatric patients, has not received the attention it deserves, and in many studies which are based on case notes the proportion of patients excluded because of incomplete data is rather large. It is not at all unlikely that these exclusions are correlated with birth order, since the family history is often based on the information provided by the patient, who is more likely to have accurate information about the ages of his younger than of his older sibs. J. A. Cobb (1914) pointed out that a patient may not even know of the existence of older siblings, especially if these died in infancy, whereas he is likely to know about younger siblings who have died. This bias would lead to an apparent excess of early-born patients.

Studies of Population Samples

After considering all these sources of bias in birth order studies, one is tempted to ask: do they really amount to anything in practice? It might seem that there are so many biases operating in so many directions that the chances are they will cancel each other out. What happens if one studies a whole population, or a random sample of one? We have already seen, in the case of the 11-year-old Scottish schoolchildren, the marked distortions of distribution by birth-rank and sibship size which occur when sibships are incomplete; and we also noted that not all the distortion could be accounted for by incompleteness of sibships, but that the excess of first-born over last-born indicated that some other bias was operating in the population. Unfortunately, no complete adult population has been studied in relation to birth order. In the case of population samples, we require that the numbers should be large, at least in the thousands, and that they should show as little selection bias as possible.

The nearest approach to a random sample of a general population is probably the random sample of employed males in the U.S.A. studied by Kohn and Schooler (in preparation). We are indebted to Dr. Schooler for some preliminary unpublished information from this sample (Table V). Out of a total of 3,091 individuals, 775 were first-born and 646 were last-born, giving a first-born–last-born ratio of 1·200; the proportion of first-born in the first- and last-born is 0·5454, and this differs from the expected proportion of 0·5 at the 0·001 level of significance. The excess of first-born is limited to family sizes two to four; the larger families show no deviation from the expected distribution. Even in families of four the distortion is probably not so marked, and it is interesting to see that there are slightly more

TABLE V

Position in sibship of a random sample of employed males in the U.S.A.; data of Kohn and Schooler

	Sibship sizes			
	1–4	5+	all	all %
First-born	561	214	775	25·1
Last-born	433	213	646	20·9
From first half of sibship (except first-born) ..	98	454	552	17·9
From last half of sibship (except last-born) ..	115	458	573	18·5
From middle of sibship (e.g. second of three) ..	183	135	318	10·3
Only children	227		227	7·3
Total sample	1,617	1,474	3,091	100·0

third-born than second-born in the families of four (115 compared with 98). It must be said that Kohn and Schooler's sibships included all children brought up in the same family as the index individuals (not only those born to the same mother), but it is difficult to see how this could account for the excess of first-born.

More plentiful, although less random, is the sample mentioned briefly by Wahl (1956). Of these 100,000 naval inductees in the U.S.A., 27 per cent. were first-born and 27 per cent. were last born. Here we find no excess of first-born, although it is possible that such an excess has been balanced by an excess of last-born due to incomplete sibships, or even by a tendency for last-born to want to go to sea.

In this country, the largest mentally healthy sample to be investigated for birth order is Norton's (1952) control group of 500 medical and surgical in-patients at a London hospital. This sample showed no deviation from random distribution by birth-rank. The numbers are comparatively small, however, and it might not be wise to generalize the finding. Moreover, since it was a metropolitan population, it is possible that an excess of first-born due to an increase in marriages was balanced by an excess of last-born due to immigration; if this was the case, we would expect the immigration effect to be greater in Norton's psychiatric group, because of the association between geographical mobility and mental illness; and in fact there was a highly significant excess of last-born among the 2,500 psychiatric patients.

CONCLUSIONS

Certain practical suggestions for the planning and reporting of birth order studies arise from the foregoing considerations. In view of the complex nature of the subject, they are presented more as a stimulus to thought than a rigid set of rules.

1. There should be an investigation of changes in the number of births and family size during the years when the sibships in question were being born, and some estimate of the effect of these changes should be made. If it is large, the method of Barker and Record (1967*b*) should be considered. The method would have to be modified for use with adult patients: perhaps the number of patients for each year of birth should be made proportional to the number of births in the parent population for that year. Another possibility is to compare the sample with a control group matched for age; however, the problems of matching for other relevant variables such as social class would probably be prohibitive.

2. Much of the bias due to the correlation between birth order and neo-natal death can

be removed by defining as siblings only those who survive the first year of life.

3. A separate analysis should be made for patients who have not migrated into the area.

4. If the mothers of any of the patients have not passed the reproductive age, care should be taken to eliminate bias due to incomplete sibships. This can be done by redefining a sibship as the children born to a mother within X years of the birth of her first child, where X is the age of the youngest patient in the sample. The sibships are thus made complete. A few patients will, of course, be lost because they are no longer members of sibships, as redefined. If the hypothesis being tested particularly concerns the last position in the sibship, then this method will not be so appropriate, as the real last-borns will be diluted by a number of technical last-borns who have not, of course, had the experience of being last-born in a family. In this case the method of Barker and Record (1967*b*) should be considered. On the other hand, Barker and Record's method requires a knowledge of the ages of all siblings, whereas for the present procedure it is only necessary to know the ages of siblings younger than the patient, and for this reason it may be more reliable with a patient population, particularly if a proportion of the family histories are not supplemented by information from one of the patient's parents.

5. No siblings or sibships should be excluded from the analysis because of death, or because further cases of the disease in question have occurred either in siblings or in more distant relatives. Death and the manifestation of disease are correlated with age, and exclusions on these grounds will lead to a deficit of patients in the later birth ranks.

6. Care should be taken in subdividing the patients in any way which might distort the random distribution of the sample between birth ranks. There may, for instance, be more last-born boys than girls in the general population. Any subdivision by sibship size will tend to exaggerate bias due to incomplete sibships or to secular changes in family size.

7. Every effort should be made to obtain data for every patient in the sample, and the number excluded on grounds of insufficient data should be stated. Of those included in the analysis, the number for whom the family history is confirmed by a member of an older generation of the patient's family should be given if possible and, if this falls far short of the whole sample, the results should be viewed with caution. The patients with family histories confirmed in this way should not be analysed separately, because the probability of confirmation is itself likely to be correlated with birth rank.

Summary

The following sources of bias in birth order studies are discussed:

1. Changes in number of births, number of marriages and family size in the general population.

2. Differential survival by birth rank, and differential migration to sources of ascertainment of patients.

3. Incompleteness of sibships.

4. Exclusions from the sample, either deliberate or because of incomplete data.

Some of the biases have opposite effects in small and large sibships, so that subdivision of the population by sibship size is likely to magnify the biases. In the British population, there is likely to be an excess of early-born in small sibships and of later-born in large sibships (due to increase in marriages and reduction in family size) and this will be even more marked in a sample whose sibships are not complete.

Some suggestions are made for the planning and reporting of birth order studies, and a method is proposed for overcoming the problem of incomplete sibships.

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