Sex Ratio in Sibships With Twins

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In national birth registers of Caucasians, the secondary sex ratio, that is, the number of boys per 100 girls at birth, is almost constant at 106. Variations other than random variation have been noted, and attention is being paid to identifying presumptive influential factors. Studies of the influence of different factors have, however, yielded meagre results. An effective means of identifying discrepancies is to investigate birth data compiled into sibships of different sizes. Assuming no inter- or intra-maternal variations, the distributions of the sex composition are binomial. Varying parental tendencies for a specific sex result in discrepancies from the binomial distribution. Over a century ago, the German scientists Geissler and Lommatzsch analyzed the vital statistics of Saxony, including twin maternities, for the last quarter of the 19th century. They considered sibships ending with twin sets. Their hypothesis was that in sibships ending with male–male twin pairs, the sex ratio among previous births is higher than normal, while in sibships with female–female twin pairs, the sex ratio is lower than normal. If the sibship ended with a male-female pair, then the sex ratio is almost normal. Consequently, a same-sex twin set indicated, in general, deviations in the sex ratio among the sibs within the sibship. Our analyzes of their data yielded statistically significant results that support their statements.

In national birth registers, the secondary sex ratio, that is, the proportion of males born, has been remarkably stable in Caucasians across time and place, varying little from a rate of 106 males per 100 females. However, nonrandom variations have been noted, and attention has thus been paid to identifying presumptive influential factors. Of special interest is the hypothesis that parents have a varying predisposition to give birth to either boys or girls. In addition, this disposition may vary depending on maternal age and birth order of the child (James, 1987a, b). Several studies have been performed to identify factors influencing the sex ratio (for references, see Discussion section).

Studies of both inter- and intra-maternal variations are difficult to conduct. Individual sibships are too small for statistical analyzes, and in official birth registers such maternal tendencies are difficult to identify. The most effective method is to investigate birth data compiled into sibships of different sizes. Assuming no maternal variations, the distribution of the sex composition of sibships is binomial. Varying maternal tendencies for a specific sex can be identified by discrepancies from the binomial distribution. However, maternal variations seem to be small, and consequently, any effect causes minute discrepancies from the theoretical distribution. Such studies therefore demand large data sets.

Geissler (1889a, 1889b) analyzed the vital statistics of the Kingdom of Saxony (in present-day Germany) mainly for the last quarter of the 19th century. He published sibship data containing almost one million sibships, and 4.8 million births grouped according to sibship size and sex composition. This data set has been the target of a long series of studies, and the results will be considered in more detail in the Discussion section.

In addition, Geissler (1896) presented and analyzed data concerning sibships ending with twin sets. He described marked variations in the sex ratio. Lommatzsch (1902, 1907), a successor of Geissler, continued the analyzes of the vital statistics from Saxony. To the best of our knowledge, the twin sibships have not been extensively investigated and therefore in this article we pay special attention to them.

Material

Geissler (1896) considered 18,283 sibships recorded in Saxony in 1881–1894, with at least three births and ending with a twin set. The total number of births before the twin sets was 82,031. Geissler collected the data from official birth cards (Zählkarte) containing information of children born earlier in the sibship. If the registered maternity was a twin pair, the birth card was extraordinarily green, and consequently, easy to identify. He noted that no information was registered concerning earlier multiple maternities in the sibship. According to Lommatzsch (1902, 1907), a

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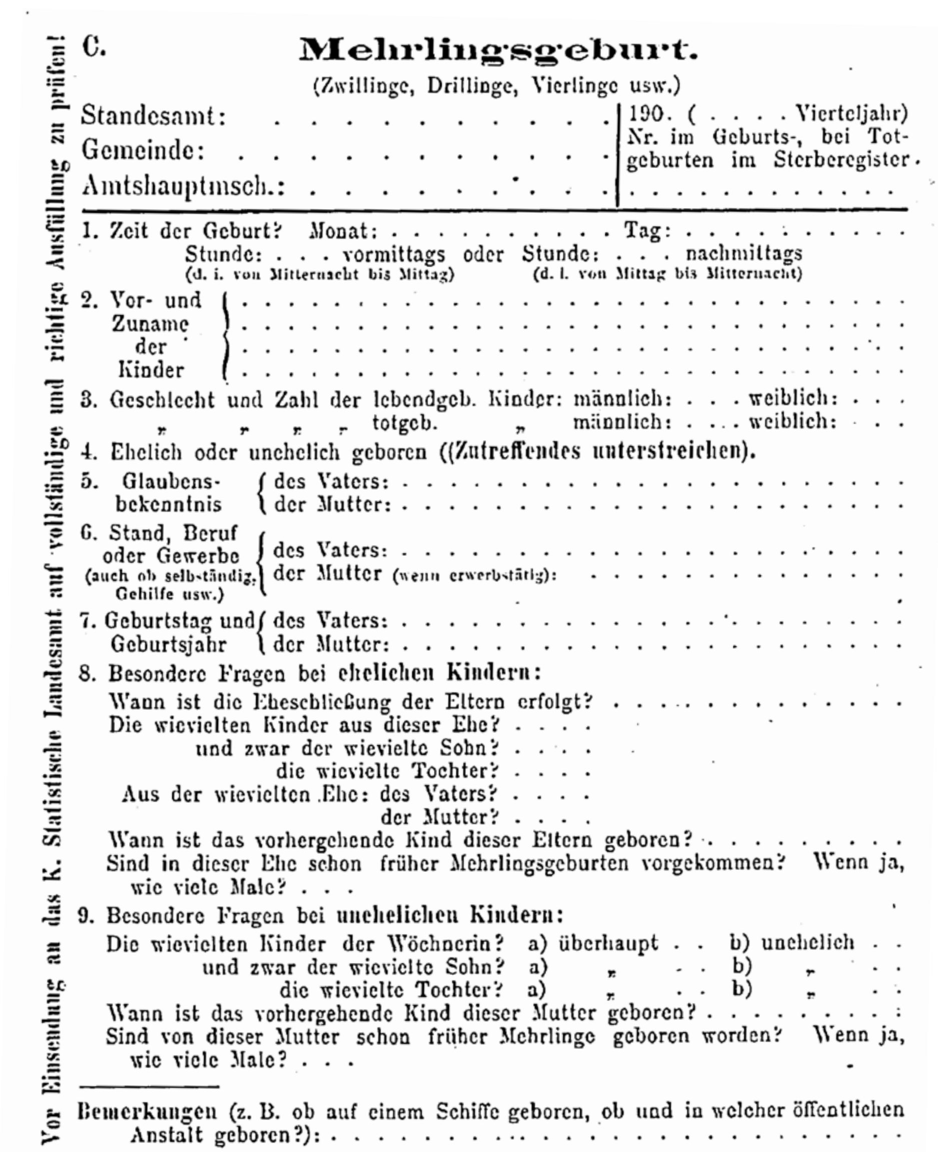
question concerning earlier multiple maternities was included in the card from 1898 onwards. Lommatzsch also published a copy of the card (Mehrlingsgeburt-Standesamtskarte) for multiple births (reprinted here in Figure 1). In this edition of the card, there was a question 'Sind in dieser Ehe schon früher Mehrlingsgeburten vorgekommen? Wenn ja, wie viele Male?' (Are there in this sibship earlier multiple maternities? If so, how many?). No information about such additional twin sets was published. In several studies, the recurrence of multiple maternities in sibships has been found to be rather common. Based on different sources, the average number of repeated multiple maternities per mother in Caucasian populations varies between 4.6% and 9.3%. This frequency is strongly associated with the twinning rate in the population (Andersen, 1980; Eriksson, 1973; Puech, 1877; Weinberg, 1901). Recently, Imaizumi (2007) reported that in Japan, with its well-known low twinning rate, the repeat frequency among couples was under 1%. In our opinion, because the data sets from Saxony are so large, and the twinning rate at that time was between 12 and 13 per 1000 maternities, additional occurrences are quite plausible.

Geissler presented the data in his tables 1a, 1b, and 1c grouped according to the three types of twin sets, male-male (MM), female-female (FF), and male-female (MF), and the size of the sibships. In Tables 1 to 3, we reprint his tables in simplified



Vor Einsendung an das K. Statistische Landesamt auf vollständige und richtige Ausfüllung zu prüfen!

C. **Mehrlingsgeburt.**

(Zwillinge, Drillinge, Vierlinge usw.)

Standesamt: . . . . | 190 . (. . . . Vierteljahr)
Gemeinde: . . . . | Nr. im Geburts-, bei Totgeburten im Sterberegister.
Amtshauptmsch.: . . . . | . . . .

1. Zeit der Geburt? Monat: . . . . Tag: . . . .
Stunde: . . . vormittags oder Stunde: . . . nachmittags
(d. i. von Mitternacht bis Mittag) (d. i. von Mittag bis Mitternacht)

2. Vor- und Zuname der Kinder . . . .

3. Geschlecht und Zahl der lebendgeb. Kinder: männlich: . . . weiblich: . . .
" " " " totgeb. " männlich: . . . weiblich: . . .

4. Ehelich oder unehelich geboren ((Zutreffendes unterstreichen).

5. Glaubensbekenntnis { des Vaters: . . . .
der Mutter: . . . .

6. Stand, Beruf oder Gewerbe (auch ob selbständig, Gehilfe usw.) { des Vaters: . . . .
der Mutter (wenn erwerbstätig): . . . .

7. Geburtstag und Geburtsjahr { des Vaters: . . . .
der Mutter: . . . .

8. Besondere Fragen bei **ehelichen Kindern:**
Wann ist die Eheschließung der Eltern erfolgt? . . . .
Die wievielten Kinder aus dieser Ehe? . . . .
und zwar der wievielte Sohn? . . . .
die wievielte Tochter? . . . .
Aus der wievielten Ehe: des Vaters? . . . .
der Mutter? . . . .
Wann ist das vorhergehende Kind dieser Eltern geboren? . . . .
Sind in dieser Ehe schon früher Mehrlingsgeburten vorgekommen? Wenn ja, wie viele Male? . . .

9. Besondere Fragen bei **unehelichen Kindern:**
Die wievielten Kinder der Wöchnerin? a) überhaupt . . b) unehelich . .
und zwar der wievielte Sohn? a) " . . b) " . .
die wievielte Tochter? a) " . . b) " . .
Wann ist das vorhergehende Kind dieser Mutter geboren? . . . .
Sind von dieser Mutter schon früher Mehrlinge geboren worden? Wenn ja, wie viele Male? . . .

Bemerkungen (z. B. ob auf einem Schiffe geboren, ob und in welcher öffentlichen Anstalt geboren?): . . . .

Figure 1

Birth card (Zählkarte) for multiple maternities according to Lommatzsch (1907). Note that this edition of the card includes the question 'Sind in dieser Ehe schon früher Mehrlingsgeburten vorgekommen? Wenn ja, wie viele Male?' [Are there in this sibship earlier multiple maternities? If so, how many?]

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Table 1
Geissler's Data (1896) for Sibships Ending With MM Twin Sets

		Male–Male (MM) twin pairs		
Children before the twin set	Sex	Number of births before twin maternity	Proportion	Number of sibships
1	Boys	592	60.22	983
	Girls	391	39.78	
2	Boys	1050	59.19	887
	Girls	724	40.81	
3	Boys	1323	54.51	809
	Girls	1104	45.49	
4	Boys	1442	51.50	700
	Girls	1358	48.50	
5	Boys	1572	54.02	582
	Girls	1338	45.98	
6	Boys	1619	52.91	510
	Girls	1441	47.09	
7	Boys	1473	53.96	390
	Girls	1257	46.04	
8	Boys	1423	55.07	323
	Girls	1161	44.93	
9	Boys	964	51.01	210
	Girls	926	49.00	
10	Boys	853	52.65	162
	Girls	767	47.35	
11 and over	Boys	1263	52.85	197
	Girls	1127	47.15	
Total	Boys	13,574	53.93	5753
	Girls	11,594	46.07	

Note: The proportions are given in percentages. A test of homogeneity in the proportion of males born before the twin set in sibships of different sizes yields $\chi^2_{10} = 53.7$, which indicates statistically significant heterogeneity.

form and translated into English. We omitted the data concerning 'births after the twin set' because these data are without additional informative value. Geissler presented all male and female births in the sibships. He admitted that a sibship, especially one of larger size, can contain triplets and/or additional twin sets. However, the estimation of the sex proportion was based on the assumption that all births before the twin set were single. A more problematic consequence of the possibility of earlier twin sets is that some late and large sibships may contain earlier smaller sibships, and accordingly, some births were counted several times and included in different lines. Statistically, this means that the actual registered number of births is lower than the number that appears in the tables. Such duplications obviously cannot be identified retrospectively. Lommatzsch (1907) continued Geissler's studies and published analogous data for the period 1901–1905. In our study, we analyze Lommatzsch's data separately. In Table 4, we present his data. Note that Lommatzsch´s data are not as detailed as Geissler's initial data.

Methods

We considered Geissler's (1896) statements about a general tendency to an excess of males or females, respectively, in sibships ending with same-sex (SS) twin pairs. We conducted a mathematical analysis concerning sibships with MM twin pairs. An analogous analysis can be applied to sibships with FF twin pairs.

The Mathematical Model

A mathematical layout of Table 1 is given in Table 5. The first two lines in Table 5 are the theoretical model and the last two lines are examples taken from Table 1. In the line i, the numbers m and f denote the total numbers of boys and girls before twin maternity for all birth orders up to and including i. The total number of sibships is denoted n.

Consider the general case that i births precede the twin set, that the number of sibships is n_i, and that the proportion of boys before the twin set is r_i. The total number of males before the twin set is $m = i\, r_i\, n_i$, and the total number of females before the twin set is $f = i(1-r_i)n_i$. In our opinion, if one is

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Table 2

Geissler's Data (1896) for Sibships Ending With FF Twin Sets

Children before the twin set	Sex	Female–Female (FF) twin pairs Number of births before twin maternity	Proportion	Number of sibships
1	Boys	364	39.14	930
	Girls	566	60.86	
2	Boys	768	44.96	854
	Girls	940	55.04	
3	Boys	1118	48.21	773
	Girls	1201	51.79	
4	Boys	1432	50.35	711
	Girls	1412	49.65	
5	Boys	1481	49.95	593
	Girls	1484	50.05	
6	Boys	1480	51.18	482
	Girls	1412	48.82	
7	Boys	1354	52.56	36
	Girls	1222	47.44	
8	Boys	1072	48.55	27
	Girls	1136	51.45	
9	Boys	887	50.54	195
	Girls	868	49.46	
10	Boys	736	50.76	145
	Girls	714	49.24	
11 and over	Boys	1303	51.60	209
	Girls	1222	48.40	
Total	Boys	11,995	49.62	5536
	Girls	12,177	50.38	

Note: The proportions are given in percentage. A test of homogeneity in the proportion of males born before the twin set in sibships of different sizes yields χ^2_{10} = 76.3, which indicates statistically significant heterogeneity.

Table 3

Geissler's Data (1896) for Sibships Ending With MF Twin Sets

Children before the twin set	Sex	Male–Female (MF) twin pairs Number of births before twin maternity	Proportion	Number of sibships
1	Boys	512	51.87	987
	Girls	475	48.13	
2	Boys	999	51.71	966
	Girls	933	48.29	
3	Boys	1505	52.48	956
	Girls	1363	47.52	
4	Boys	1901	52.51	905
	Girls	1719	47.49	
5	Boys	2064	51.79	797
	Girls	1921	48.21	
6	Boys	1980	51.97	635
	Girls	1830	48.03	
7	Boys	1843	50.44	522
	Girls	1811	49.56	
8	Boys	1616	52.33	386
	Girls	1472	47.67	
9	Boys	1410	50.21	312
	Girls	1398	49.79	
10	Boys	1096	50.51	217
	Girls	1074	49.49	
11 and over	Boys	1958	51.95	311
	Girls	1811	48.05	
Total	Boys	16,884	51.65	6994
	Girls	15,807	48.35	

Note: The proportions are given in percentage. A test of homogeneity in the proportion of males born before the twin set in sibships of different sizes yields χ^2_{10} = 8.39. This value is not statistically significant and homogeneity can be accepted.

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Table 4

Lommatzsch's Data (1907) for Sibships Ending with MM, FF, and MF Twin Sets

Children before the twin set	Sex	Number births before twin maternity	Proportion	Number of sibships
		1a (MM) twin pairs		
1–3	Boys	282	61.04	462
	Girls	180	38.96	
4–6	Boys	1572	55.35	979
	Girls	1268	44.65	
7–9	Boys	1656	55.09	513
	Girls	1350	44.91	
10–12	Boys	1174	54.00	248
	Girls	1000	46.00	
11–13 and over	Boys	809	52.98	125
	Girls	718	47.02	
Total	Boys	5493	54.88	2327
	Girls	4516	45.12	
		1b (FF) twin pairs		
1–3	Boys	156	38.61	404
	Girls	248	61.39	
4–6	Boys	1252	46.30	933
	Girls	1452	53.70	
7–9	Boys	1427	49.55	493
	Girls	1453	50.45	
10–12	Boys	1061	50.55	241
	Girls	1038	49.45	
11–13 and over	Boys	616	50.41	91
	Girls	606	49.59	
Total	Boys	4512	48.47	2162
	Girls	4797	51.53	
		1c (MF) twin pairs		
1–3	Boys	255	54.72	466
	Girls	211	45.28	
4–6	Boys	1820	51.88	1192
	Girls	1688	48.12	
7–9	Boys	2139	51.84	701
	Girls	1987	48.16	
10–12	Boys	1567	51.33	349
	Girls	1486	48.67	
11–13 and over	Boys	868	52.35	134
	Girls	790	47.65	
Total	Boys	6649	51.90	2842
	Girls	6162	48.10	

Note: Geissler's tables have been taken as a model for this table. The proportions are given in percentage. A test of homogeneity in the proportion of males in sibships of different sizes yields significant values for MM and FF cases, but not significant values for MF cases.

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Table 5

Mathematical Model For an Observed Example of Geissler's Data

Children before the twin set	Sex	MM twin pairs Number of births before twin maternity	Proportion	Number of sibships
i	Boys	m	$\frac{m}{m+f}$	n
	Girls	f	$\frac{f}{m+f}$	
3	Boys	1323	55.51	809
	Girls	1104	45.49	

Note: Numerical data are extracted from Table 1.

interested in tracing the tendencies of extreme sex ratios, then calculations must be based on data from before the twin sets.

Results

Analyzes of Sibship Data

In Figure 2, we present the proportion of males born before the twin sets in Tables 1 to 3. If one investigates the data in Tables 1 and 2 and Figure 2, the proportion of males shows rather strong fluctuations starting from extreme levels, but after that converges to levels closer to normal. However, for sibships ending with MM twin sets, the proportion of males is, in general, too high, while for sibships ending with FF twin sets, the proportion is too low. For sibships ending with MF twin sets, the proportion of males shows only small fluctuations and is normal.

In the class of sibships with three children, including a MM twin set (Table 1), there are 592 boys and 391 girls. Consequently, these 983 sibships with three children contain 592 (60.2%) all-male sibships. The twin sets can be MZ or DZ, so one cannot decide if there are two or three fertilizations resulting in three males. According to the corresponding analysis of the class of sibships with three children, including a FF twin set (Table 2), there are 566 girls and 364 boys. Consequently, these 930 sibships with three children comprise 566 (60.9%) all-female sibships. Still, it cannot be determined whether there are two or three fertilizations resulting in three females. The proportions of males and females obtained in the class of sibships with three children in Table 3 are rather normal.

We consider the whole data sets in Tables 1 to 3 and test the homogeneity in the proportion of males before the twin sets in sibships of different sizes. These

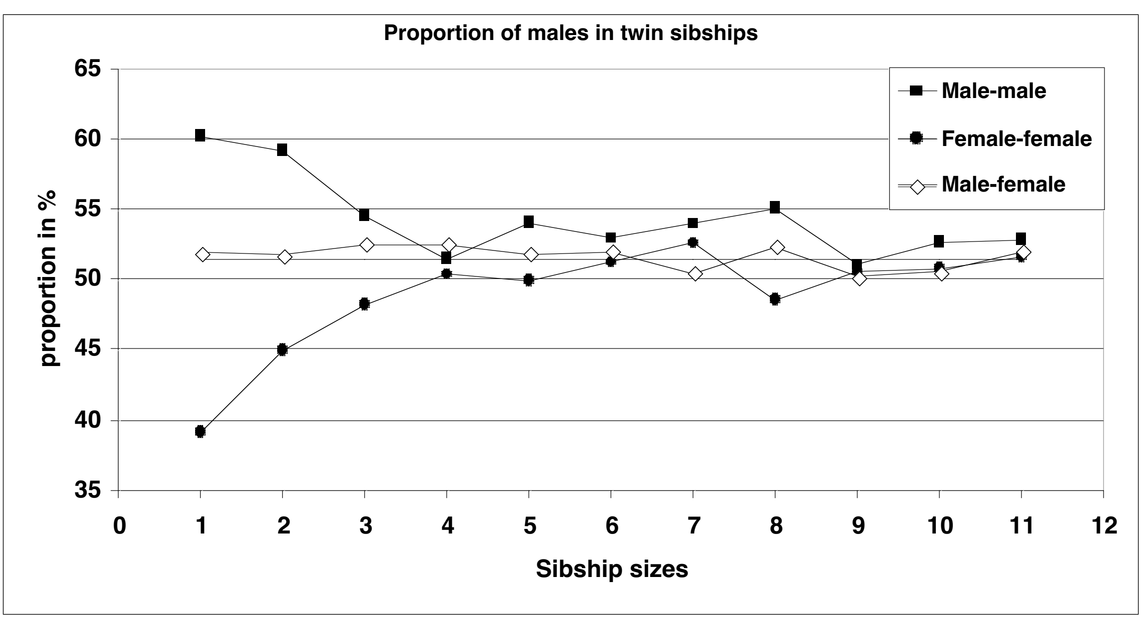

Figure 2

Proportion of males before the twin sets. The proportions converge with increasing sibship sizes. However, for male–male twin sibships, the proportion of males is, in general, slightly too high, and for female–female sibships slightly too low. The 'normal' proportion (51.5%) is indicated in the figure.

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tests are based on the assumption that the data in the rows are independent, that is, no small sibships are included in later larger sibships with recurrent twinning. For different sibship sizes with MM twin sets, we obtain χ^2_{10} = 53.7 with ten degrees of freedom. This value is statistically significant. For FF twin sets, we obtain χ^2_{10} = 76.3, which is also statistically significant. The contributions to the high χ^2_{10} values in these two cases are mainly caused by the cases i = 1 and 2. These extreme proportions are discernible in Figure 2. In sibships with MF twin sets, variation in the proportions is not significant (χ^2_{10} = 8.39).

If we consider Lommatzsch's data in Table 4 and test the homogeneity in the proportion of males born before the MM twin sets for different sibship sizes, we obtain χ^2_4 = 29.2, which is statistically significant. For FF twin sets, we obtain χ^2_4 = 89.2, which is also statistically significant. The contributions to the high χ^2_4 values are mainly caused by the case i = 1. In sibships with MF twin sets, variation in the proportions is not significant (χ^2_4 = 3.60).

The total sums of births before the twin sets in Geissler's data are given in Table 6. Included in the table are the proportions of males and females and the SEs of these proportions. If we test the proportions obtained for the total numbers in the three groups of sibships (MM, FF, and MF) against each other, we obtain χ^2_2 = 92.0, with two degrees of freedom, yielding p = < .001. This result indicates statistically significant differences between the sex ratios for the three groups. If we consider Lommatzsch's data in Table 7 and test the proportions obtained for the three groups of sibships against each other, we obtain χ^2_2 = 79.4, with two degrees of freedom, yielding p = < .001.

Table 6

Total Numbers of Males and Females Before the Twin Set in Sibships With MM, FF and MF Twin Sets According to Geissler's Data

Twin set	Males	Females	Total	SE
Male–male	13,574	11,594	25,168	
Proportions, %	53.93	46.07		0.314
Female–female	11,995	12,177	24,172	
Proportions, %	49.62	50.38		0.322
Male–female	16,884	15,807	32,691	
Proportions, %	51.65	48.35		0.276
Total	42,453	39,578	82,031	
Proportions, %	51.75	48.25		0.175

Note: The SEs of the estimated proportions are the same for both males and females. If we test the proportions obtained for the three groups of sibships (MM, FF, and MF) against each other, we obtain χ^2_2 = 92.0, with two degrees of freedom, yielding p < .001.

Table 7

Total Numbers of Males and Females Before the Twin Set in Sibships with MM, FF, and MF Twin Sets According to Lommatzsch's Data

Twin set	Males	Females	Total	SE
Male–male	13,574	11,594	25,168	
Male–male	5493	4516	10,009	
Proportions, %	54.88	45.12		0.497
Female–female	4512	4797	9309	
Proportions, %	48.47	51.53		0.518
Male–female	6649	6162	12,811	
Proportions, %	51.90	48.10		0.441
Total	16,654	15,475	32,129	
Proportions, %	51.83	48.17		0.279

Note: The SEs of the estimated proportions are the same for both males and females. If we test the proportions obtained for the three groups of sibships (MM, FF, and MF) against each other, we obtain χ^2_2 = 79.4, with two degrees of freedom, yielding p < .001.

Brown et al. (2001) stated that at realistic and even larger than realistic sample sizes the actual coverage probability of the standard confidence interval (CI) for a proportion can differ markedly from the nominal confidence level. They stressed that the CIs presented by Wilson (1927) and Agresti and Coull (1998) give the best consistency between actual and nominal confidence levels. However, when we compared the different CIs very minute differences were obtained. The explanation for this is that the standard CI is rather robust, when the proportion is close to 50%. Therefore, we chose to use the standard CI here. For the MM sibships in Table 1, the 95% CI for the proportion of boys is 53.3–54.6. This CI excludes the 'normal' value of 51.5%. For the FF sibships in Table 2, the 95% CI for the proportion of boys is 49.0–50.2, which also excludes the 'normal' value. For the MF sibships in Table 3, the 95% CI for the proportion of boys is 51.1–52.2. This CI includes the 'normal' value of 51.5%.

Similar results were obtained for the Lommatzsch data. For the MM sibships, we found the CI 53.9–55.9, for the FF sibships 47.4–49.5, and for the MF sibships 51.0–52.8. Only the last CI includes the 'normal' value of 51.5%. The Lommatzsch data thus support the findings of the Geissler data.

Remark

The SEs of mean proportions obtained for heterogeneous groups are less than the SEs for homogeneous ones (Fellman & Eriksson, 2002). Consequently, in our study, the CIs are too long, and the strengths of the significance are less than if heterogeneity had been considered. Our tests and CIs, based on the assumption that the proportions are independent of the sibship size, are therefore conservative.

Discussion

Geissler's (1889a, b) data. Lancaster (1950) considered Geissler's data and analyzed the proportion of males in different sibships. He distinguished between complete sibships of eight individuals and sibships before the ninth birth. He observed intra-family varia-

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tions. His main conclusion was that Geissler's data are biased, probably because of careless answers by parents, and that definite conclusions cannot be drawn from this data to support the theory that heterogeneity exists in the probability of a birth being a boy between sibships.

Fisher (1954, pp. 66–68) analyzed, as an example, Geissler's data concerning the distribution of boys and girls in sibships of size 8. He found an overdispersion, which indicates interfamilial heterogeneity. One argument for this could be the occurrence of identical twin maternities. However, he observed that the frequency of twin maternities was insufficiently large to be the only cause.

Inspired by Fisher's studies of Geissler's data, Edwards (1958) applied the generalized binomial distribution, with a parameter which is beta-distributed within the population. He concluded that the probability of a birth being male varies between families of the same size, and that there is no evidence supporting the existence of parents only capable of producing unisexual families.

Later, Edwards (1962) again considered Geissler's data and gave a thorough presentation of earlier studies on these data. According to Edwards, concern regarding the quality of the data set varies. Furthermore, Edwards suggests different methods to trace intra- and interfamily variations. For a survey of additional early references, see Edwards (1962).

Lindsey and Altham (1998) also paid much attention to Geissler's data. They analyzed the data by building alternative stochastic models (mainly the beta-binomial, the 'multiplicative' binomial and the double-binomial models). Furthermore, they stressed that two main questions emerge for the sex ratio. Does the probability for a boy between families, and/or within a family, vary over time? These alternatives cannot be distinguished because they will both manifest themselves as overdispersion with respect to the binomial model. Thus, the second question one can ask is whether the variability changes with the size of the family; if it does, it could be a result of either or both of the factors mentioned. Finally, they remark that there is common agreement that the overdispersion cannot be explained by twin or other multiple maternities. Lindsey and Altman (1998) found the best fit when they assumed an increasing sex ratio and a decreasing overdispersion with increasing sibship size. They also estimated the probability of couples capable of only having children of one sex. The estimated probability was small (.000246), indicating about 250 families of this kind in the data set. This number is remarkably low compared with Geissler's data, where there are 191,634 (19.2%) unisexed sibships. This result should be compared with Edwards´ statement that he could not identify such families. In a later broad survey, Lindsey (1999) continued to discuss these findings. Recently, Stansfield and Carlton (2007) studied the sex ratio in two- and three-child sibships. They observed statistically significant discrepancies from the binomial model, and could identify the effect of family planning on the distribution of boys and girls within the sibships. In fact, Gini (1951) had already discussed the effect of family planning on the sex ratio within sibships.

Geissler's (1896) data. Geissler's own conclusions concerning these data were that in sibships ending with MM twin pairs, the sex ratio was markedly higher than normal. Analogously, he also noted that in sibships ending with FF twin pairs the sex ratio was extremely low. Furthermore, he noted that in sibships ending with MF twin pairs the sex ratio was normal. He concluded that his findings speak in favour of a general tendency to an excess of males or females, respectively, in sibships where SS twin pairs are found. His successor Lommatzsch (1907) agreed with this conclusion.

Weinberg (1901, p. 358) analyzed Geissler's material from 1896 in a different way. He was interested in the sex ratio among the twins following a single male or a single female. He observed that after a single male, the proportion of males among the twins increased, to a rate of 57.8% (the value of 58.1% presented by Weinberg is slightly incorrect). After a single female, Weinberg noted a decreased proportion of males among the twins, giving a (correct) value of 43.9%. The proportion of males among all twins was 50.9% (50.8% according to Weinberg). This sex proportion was rather low, but the proportion of males among the firstborn singletons was also low, only 50.6%.

However, Weinberg felt dubious about Geissler's results and tried to confirm them by studies of twin and triplet families, including Geissler's and his own material. These attempts did not give supporting results. Weinberg's only explanation for discrepancies between the series was that Geissler's data were based on personal interviews and not on official records, and consequently, were not reliable enough.

Wedervang (1924) also analyzed Geissler's sibship data in detail. He concentrated his analyzes on the data before the twin sets. He noted a slight increase in the sex ratio, but was critical of the data. According to Wedervang, insufficient information exists on the births before the twin sets. These births might be twin sets, and if they are SS twins their influence on the sex ratio can be marked. However, such additional SS twin sets of the same type may support a tendency for a specific sex. Against Wedervang's remarks is that the most extreme sex ratios were obtained for the group '1'. In this group, no SS twin sets occurred before the index set.

Figure 3 presents the proportions of MM, FF, and MF twin sets for different sibship sizes. With increasing sibship sizes, the mean maternal age for the index twin set increases. Hence, the increasing proportion of MF sets is a consequence of the association between maternal age and sibship size, and the

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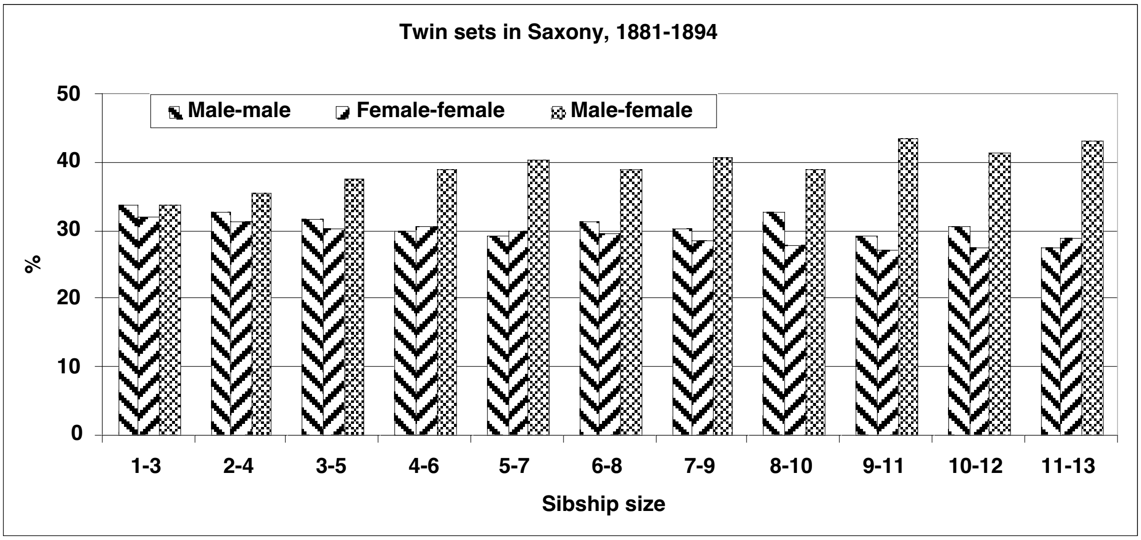

Figure 3

Proportions of MM, FF, and MF twin sets among the index sets for different sibship sizes. With increasing sibship sizes, the maternal age for the index twin set increases. The increasing proportion of MF sets is a consequence of the association between maternal age and sibship size (parity) and the increasing DZ twinning rate with increasing maternal age.

increasing DZ twinning rate with increasing maternal age. All of our analyzes of the sex ratio based on the data before the twin sets indicate that in MM sibships the observed proportion of males, and in FF sibships the observed proportion of females, are slightly, but significantly higher than normal. Hence, based on the published data, our conclusion is in agreement with the statements by Geissler and Lommatzsch that SS twin sets show a slight tendency for deviations in the sex ratio. Finally, it is necessary to give some additional criticism of Geissler's (1896) data sets. The problem with personal interviews has already been discussed. An apparently more important shortcoming of the data sets is that no information about the distributions of males and females within the sibships is available.

Factors influencing the sex ratio. In a long series of papers, attempts have been made to identify factors influencing the sex ratio within families. There has been a widespread belief that the sex ratio at birth varies inversely with the frequency of prenatal losses. However, the available data on late fetal mortality lend at best only weak support for this hypothesis (Visaria, 1967). Increases in the sex ratio at birth have been reported for some human malformations (Arena & Smith, 1978) and in some parental disorders, for example, toxaemia of pregnancy, mothers with multiple sclerosis or placenta previa, and fathers with prostatic cancer, and furthermore during warfare, and in children conceived during the summer season (James, 1987a, b). Other variables reported to be associated with an increase in the secondary sex ratio are large family size, high ancestral longevity, paternal baldness, excessive coffee-drinking, intensive coital frequency, and illegitimacy (Teitelbaum, 1972). Biased sex ratios have also been noted in connection with neural tube defects; in general, the sex ratio was low, but for low spinal lesions, particularly those in the sacrum, the sex ratio showed an excess of males (Seller, 1987). James (1987b, 1996) has hypothesized that the hormone levels of both parents at the time of conception affect the probability of a male birth, with high levels of oestrogen and testosterone increasing this probability, and high levels of gonadotrophin decreasing it.

Genetic control of the sex ratio at birth could occur either through control of the primary sex ratio or through subsequent differential sex mortality. The only clear cut case of distortion of segregation, or meiotic drive, in man involves the D-21 translocation, which is implicated in a certain proportion of cases of Down's syndrome. Normal carrier males rarely transmit the D-21 combination to their offspring (only about 5% of that expected). This example clearly shows that genetic variation can affect the segregation ratio. Some striking examples can be found in the literature of unisexual pedigrees extending over several generations, which, if valid, suggest the existence of sex-ratio genes like those reported in Drosophila (see Fellman et al., 1999; Stern, 1960). Slater (1943) stated that aberrant sex ratios tend, to a slight extent, to run in families, but to our knowledge no further studies have been carried out to confirm this observation. The finding by Lindsey and Altman (1998) that the probability of couples being only capable of having children of one sex is very low supports Slater's statement.

There may be a disturbance in the primary sex ratio, possibly due to meiotic drive (primary

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segregation disturbance or genetic selection), resulting in overproduction or preference of the egg for Y sperm. Some indications for this come from the observation of an unusually high proportion of males in Koreans (somewhat above 0.53; sex ratio about 113). However, Korean mothers have a normal sex ratio among their progeny, ruling out effects of sex-differential mortality in utero (Morton et al., 1967).

The variation in the sex ratio that has been reliably identified has always been slight, as compared with what we have noted in X-linked recessive retinoschisis (cleavage of retinal layers). With the methods used in the 1960s, we noted a marked excess of males within families with X-linked retinoschisis, but normal sex ratios in the X-linked recessive disorders haemophilia and colour blindness (Eriksson et al., 1967). However, with the exception of the X-linked recessive retinoschisis data expanded and tested with more advanced statistical methods (Fellman et al., 1999, 2002; Huopaniemi et al., 1999), there are no unequivocal examples of genes in man that affect the sex ratio. Summing up, to the best of our knowledge, the only genetic characteristic observed to have a marked influence on the sex ratio within families is X-linked retinoschisis.

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