Stillbirths in Multiple Births: Test of Independence

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The stillbirth rate in twins is a more sensitive indicator of environmental hazards than the stillbirth rate in singletons. Medical care or other socioeconomic factors may be more influential for perinatal survival in twin than in single deliveries. Studies have indicated that stillbirths among children in a set of multiple maternities are not independent. Models were considered assuming independent outcomes within a set of multiple maternities. Analyses of the stillbirth rates confirm that the risk of stillbirth among males is almost constantly higher than among females. Any model introduced should assume different stillbirth rates for males and females. The models were tested with both maximum likelihood and minimum χ^2 methods. Data was analyzed from Sweden, the Åland Islands, Saxony, England and Wales, and significant discrepancies obtained from the independence models. The same-sexed twin data contain both monozygotic and dizygotic twin sets with apparently different stillbirth rates. Consequently, for same-sexed twins the proposed model could be considered too simple. After improvement by splitting the same-sexed data into monozygotic and dizygotic twin sets, the dependence still remains. The proportion of both same-sexed and opposite-sexed twin pairs that contain two stillborn is greater than what the stillbirth rates and the independence should indicate. Consequently, stillbirth rate estimates based on the relative frequency of twin sets with two stillborn children have a positive bias. When the stillbirth rate decreases, the number of sets with two stillborn children decreases more slowly than would be indicated by independence.

The stillbirth rate (SBR) in twins is a more sensitive indicator of environmental hazards than the stillbirth rate in singletons (Boklage, 1987; Källén, 1991; Lommatzsch, 1902; Wedervang, 1924). Consequently, variations in medical care or other socioeconomic factors may be more influential to perinatal survival in twin than in singleton deliveries. Studies of offspring in multiple maternities have indicated that stillbirths among the children in a set of multiple maternities are not independent (Salihu et al., 2004). The aim of this study was to identify the effects of such dependence using stochastic models based on the assumption of independence. Preliminary studies of the effect of different SBRs for monozygotic (MZ) and dizygotic (DZ) twins were also performed, but the effect of zygosity will be discussed in more detail in a forthcoming paper (Fellman & Eriksson, 2006a). Analyses of the SBRs for different types of twin pairs confirm that the liability to stillbirth is almost constantly higher among males than among females (Fellman & Eriksson, 2006b; Eriksson & Fellman, 2006). Consequently, in the models introduced, we assumed different SBRs for males and females. The models were tested with both the maximum likelihood (ML) and the minimum χ^2 methods.

Material

We analyzed data compiled by the authors from Sweden (1869–1967). Comparisons were performed with data from England and Wales (1996–2003) obtained from the website http://www.statistics.gov.uk/, from Saxony (1881–1900) published by Lommatzsch (1902), from the Åland Islands (1750–1949) published by Eriksson (1973) and with data from England and Wales (1938–1948) earlier published by Lowe and Record (1951).

Our Swedish series starts with birth data for the period 1869 to 1878, published by Berg (1880), where exactly observed numbers were given for the composition of twin pairs and triplet sets with respect to live-births or stillbirths and to sex combinations. Similar information published by Statistics Sweden was available for Sweden for the period 1901 to 1967. Swedish data have also been considered by Fellman and Eriksson (2006b) and Eriksson and Fellman (2006). In Fellman and Eriksson (2006c), the variation in the secondary sex ratio was considered among singletons and twins, and the frequencies of MZ and DZ twin maternities and the reliability of Weinberg's differential rule estimated. In Fellman and Eriksson (2006b), the temporal variations in the SBR were considered among singletons, twins and triplets. In that study

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the temporal trends only were of interest, and not the influence of additional factors such as marital status, urbanisation or the sex combination of multiple births. Therefore, the whole period 1869 to 2001 could be considered. Eriksson and Fellman (2006) also included influential factors and the analyses were based on Swedish data for the period 1869 to 1967.

Lommatzsch (1902) analyzed data from Saxony, 1876 to 1900, in detail. Especially, he presented multiple birth data for the period 1881 to 1900 containing information about the sex combination and the condition at birth (live or still).

Eriksson (1973) presented twin data from Åland, 1653 to 1949. We included this data for the period 1750 to 1949 in the present study. The condition at birth and the sex combination of the twin sets are crucial and consequently, in our opinion, the data before 1750 contain too many cases with unknown sex and condition at birth. Furthermore, the registered number of twin sets indicates that before 1750 there may be missing data.

Lowe and Record (1951) presented the distribution of twin sets for England and Wales (1938–1948) according to the number of stillborn children. These data, especially Table 5, are also analyzed by the authors. However, Lowe and Record published data are not as informative as the authors' data from Sweden, the Åland Islands and England and Wales, or Lommatzsch's data from Saxony. Using the percentages presented by Lowe and Record (1951), the numbers of twin sets were recalculated by the authors in the different classes. A more restrictive limitation in Lowe and Record data was that they did not distinguish between males and females. Consequently, our analyses of Lowe and Record data are based on the additional assumption that the SBRs are the same for males and females.

Methods

Dependence Among the Stillbirths in a Twin Set

It is obvious that the events of live-birth and stillbirth among the individuals in a multiple birth set are dependent (Boklage, 1987; Källén, 1991; Salihu et al., 2004; Wedervang, 1924). In this study, we investigated the dependence by applying stochastic models. We assumed the null hypothesis that the outcomes of two twins in a twin set are independent and that the probability of a stillborn male is p and of a stillborn female q. When these probabilities are multiplied by 1000, one obtains estimates of the SBRs. We first considered the data of same-sexed (SS) twin pairs. We analyzed the male–male case in detail, and the analysis of the female–female case was similar.

Assume that the number of twin sets with two live-born is n_{11}, with one live-born and one stillborn is n_{12}, with two stillborn is n_{22} and that $n = n_{11} + n_{12} + n_{22}$. Starting from the independence model in Table 1, we obtain the likelihood function

$$L(p) = \left[(1-p)^2\right]^{n_{11}}\left[2p(1-p)\right]^{n_{12}}\left(p^2\right)^{n_{22}} = Cp^{2n_{22}+n_{12}}(1-p)^{2n_{11}+n_{12}}$$

and the log-likelihood function

$$l(p) = (2n_{22}+n_{12})\ln(p) + (2n_{11}+n_{12})\ln(1-p).$$

We obtain

$$\frac{dl}{dp} = \frac{2n_{22}+n_{12}}{p} - \frac{2n_{11}+n_{12}}{1-p}$$

and

$$\frac{d^2l}{dp^2} = -\frac{2n_{22}+n_{12}}{p^2} - \frac{2n_{11}+n_{12}}{(1-p)^2}.$$

The equation $\frac{dl}{dp} = 0$ gives the maximum likelihood estimator (MLE)

$$\hat{p} = \frac{n_{12}+2n_{22}}{2n},$$

that is, the relative frequency of a stillborn among all twins. Still assuming independence, we estimate

$$-E\left(\frac{d^2l}{dp^2}\right) = \frac{2n}{p(1-p)}$$

and obtain

$$Var(\hat{p}) = \frac{p(1-p)}{2n}.$$

Consequently, using the notations in Table 2 we obtain for the female–female case

$$\hat{q} = \frac{n_{12}+2n_{22}}{2n} \quad \text{and} \quad Var(\hat{q}) = \frac{q(1-q)}{2n}.$$

Consider the model in Table 3 for opposite-sexed (OS) pairs. We obtain the likelihood function

$$L(p) = \left[(1-p)(1-q)\right]^{n_{11}}\left[q(1-p)\right]^{n_{12}}\left[p(1-q)\right]^{n_{21}}(pq)^{n_{22}}$$
$$= (1-p)^{n_{11}+n_{12}}(1-q)^{n_{11}+n_{21}}p^{n_{22}+n_{21}}q^{n_{22}+n_{12}}$$

and the log-likelihood function is

$$l(p) = (n_{22}+n_{21})\ln(p) + (n_{11}+n_{12})\ln(1-p) + (n_{22}+n_{12})$$
$$\ln(q) + (n_{11}+n_{21})\ln(1-q).$$

The derivatives are

$$\frac{\partial l}{\partial p} = \frac{n_{22}+n_{21}}{p} - \frac{n_{11}+n_{12}}{1-p} \quad \text{and} \quad \frac{\partial l}{\partial q} = \frac{n_{22}+n_{12}}{q} - \frac{n_{11}+n_{21}}{1-q}.$$

The second derivatives are

$$\frac{\partial^2 l}{\partial p^2} = -\frac{n_{22}+n_{21}}{p^2} - \frac{n_{11}+n_{12}}{(1-p)^2}, \quad \frac{\partial^2 l}{\partial p \partial q} = 0$$

and

$$\frac{\partial^2 l}{\partial q^2} = -\frac{n_{22}+n_{12}}{q^2} - \frac{n_{11}+n_{21}}{(1-q)^2}.$$

The equations

$$\frac{dl}{dp} = \frac{dl}{dq} = 0$$

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give the MLEs

$$\hat{p} = \frac{n_{21} + n_{22}}{n} \quad \text{and} \quad \hat{q} = \frac{n_{12} + n_{22}}{n},$$

that is, the relative frequencies of stillborn male and female twins, respectively. In order to obtain the variances we estimate

$$\left(\frac{\partial^2 l}{\partial p^2}\right) = \frac{n}{p(1-p)} \quad \text{and obtain} \quad Var(\hat{p}) = \frac{p(1-p)}{n}$$

and analogously from

$$-E\left(\frac{\partial^2 l}{\partial q^2}\right) = \frac{n}{q(1-q)} \quad \text{the variance} \quad Var(\hat{q}) = \frac{q(1-q)}{n}.$$

Note that

$$-E\left(\frac{\partial^2 l}{\partial p \partial q}\right) = 0$$

and consequently, the estimators are uncorrelated and $Cov(\hat{p}, \hat{q}) = 0$.

Minimum χ^2 Estimation

In the Results section we observe that the proposed model does not fit the data. Especially, we note that, measured with the contribution to the χ^2 value, the frequency of sets with two stillborn children diverges most strongly from the proposed model. An alternative method of fitting the data to the model is to use the minimum χ^2 estimation. This method minimizes the function

$$\chi^2 = \sum_i \frac{(n_i - n_i(p))^2}{n_i(p)}$$

where n_i is the observed number, $n_i(p)$ is the expected number according to the proposed model and p is the unknown parameter(s). This procedure yields the absolute minimum for the χ^2, which indicates that a better fit, measured with the χ^2 test, cannot be obtained with any other method. When the models hold, the MLEs and the minimum χ^2 estimates are asymptotically equally efficient. However, we prefer the MLEs, because they equal the relative frequency of stillborn, that is, if they are given in per 1000, they are identical to the traditional SBRs.

The Effect of Zygosity

One possible factor causing the failure to fit the independence model is that the SS twin series consists of both MZ and DZ twins. If the SBRs among the MZ and DZ twins are different, then the formula np^2 for the expected number of sets with two stillborn does not hold.

We analyze the effect of zygosity, using the following notations:

n_1 is the number of MZ (male–male) twin sets
p_1 is the SBR among the MZ twins
n_2 is the number of DZ (male–male) twin sets
p_2 is the SBR among the DZ twins and $p_1 = p_2 + \delta$ ($\delta \neq 0$).

For the total series, we introduce the notations $n = n_1 + n_2$ and

$$p = \frac{n_1 p_1 + n_2 p_2}{n_1 + n_2}.$$

If we do not know or if we ignore the division into MZ and DZ sets, the expected number of twin sets with two stillborns is still np^2. However, if we consider the division into MZ and DZ twin sets, the expected number of twin sets with two stillborns is $n_1 p_1^2 + n_2 p_2^2$. In order to estimate the effect of the information about MZ and DZ twin sets, we consider the difference

$$n_1 p_1^2 + n_2 p_2^2 - n\left(\frac{n_1 p_1 + n_2 p_2}{n}\right)^2 = n_1 p_1^2 + n_2 (p_1 + \delta)^2 -$$

$$n\left(\frac{n_1 p_1 + n_2 (p_1 + \delta)}{n}\right)^2 = \quad n_1 p_1^2 + n_2 \left(p_1^2 + \delta^2 + 2 p_1 \delta\right) -$$

$$n\left(\frac{n_1 p_1 + n_2 p_1 + n_2 \delta}{n}\right)^2 = np_1^2 + n_2 \left(\delta^2 + 2 p_1 \delta\right) - \frac{(np_1 + n_2 \delta)^2}{n}$$

$$= np_1^2 + n_2 \left(\delta^2 + 2 p_1 \delta\right) - \frac{n^2 p_1^2 + n_2^2 \delta^2 + 2 n n_2 p_1 \delta}{n}$$

$$= \frac{n n_2 \delta^2 - n_2^2 \delta^2}{n} = \frac{n_2 (n - n_2)}{n} \delta^2 \geq 0.$$

Note that, independently of the sign of δ, this difference is always positive. Consequently, if one includes in the model the division of the series into MZ and DZ sets, then the expected number of sets with two stillborn is slightly increased. Hence, the ratio between the observed and the expected is somewhat decreased. However, in the Results section we observe that this improvement can reduce the high ratio values only to a small degree. Furthermore, the improvement of the model for SS twins reduces the χ^2 values only to a small extent. Consequently, the discrepancies with respect to the model are still statistically significant. In addition, this attempt cannot reduce the high ratio values for OS twins. In a forthcoming paper (Fellman & Eriksson, 2006a) we analyze the SBR among MZ and DZ twins in more detail.

Sets of Triplets and Higher Multiple Maternities

The statistical ideas discussed above can be generalized to higher multiple maternities. Assume still independence and that the probability of a stillborn male is p and of a stillborn female is q. Consider a k-tuplet maternity containing u live-born males, v live-born females, r stillborn males and s stillborn females, that is $u + v + r + s = k$. The probability for such a k-tuplet is

$$P_{u,v,r,s} = \frac{k!}{u!\,v!\,r!\,s!} (1-p)^u (1-q)^v p^r q^s.$$

Within this framework we can consider all types of multiple maternities. However, the data sets for twins are often large enough for more detailed and more informative methods. On the other hand, for higher

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multiple maternities the data are usually so rare that reliable results based on this model seem to be unobtainable. Consequently, in our opinion, this method is mainly applicable to triplet data.

Starting from the probability distribution introduced above, we can build a likelihood function and obtain MLEs for p and q. Skipping detailed mathematical steps, which are analogous with the derivations presented above, we note that for higher multiple maternities the estimates are

$$\hat{p} = \frac{\text{number of stillborn males}}{\text{total number of males}}$$

and

$$\hat{q} = \frac{\text{number of stillborn females}}{\text{total number of females}}.$$

The corresponding variances are

$$Var(\hat{p}) = \frac{p(1-p)}{kn} \text{ and } Var(\hat{q}) = \frac{q(1-q)}{kn}$$

respectively.

Although this model is, in practice, applicable to triplet data, a simpler control of the independence can also be suggested. Consider the probability that all the children in the k-tuplet are stillborn. If we note that $u = 0$ and $v = 0$, the probability of this case is

$$\sum_{r,s,r+s=k} P_{0,0,r,s} = \sum_{r,s,r+s=k} \frac{k!}{r!\,s!} p^r q^s = (p+q)^k$$

The probabilities p and q can be estimated using the whole triplet data set. After that, the estimated probability $(\hat{p} + \hat{q})^k$ can be compared with the observed relative frequency of sets with only stillborn children.

Results

The independence models and the observed data from Sweden are given in Tables 1, 2 and 3. In the tables are also given the MLEs, $\hat{p}$ and $\hat{q}$ and the χ^2 values, which have one degree of freedom. According to the χ^2 tests, the models do not fit the empirical data, and the null hypothesis (independence) has to be rejected. Tests of the models based on parameters estimated by the minimum χ^2 method yield somewhat lower χ^2 values, but still statistically significant results.

The strongest discrepancy, measured with the contribution to the χ^2 value, is the number of sets with two stillborn children. Consequently, we pay this discrepancy special attention. If we assume that the models hold, then the expected number of male–male twin sets with two stillborn males is np^2, female–female twin sets with two stillborn females is nq^2 and male–female twin sets with a stillborn male and a stillborn female is npq. We combine the two parameters in the male–female case to one argument by introducing the geometric mean $\tilde{p} = \sqrt{pq}$. Consequently, the expected number of twin sets with two stillborn is $npq = n\tilde{p}^2$. The number n is the total number of the corresponding type of twin set. In Figure 1a and 1b we present the ratio between the observed number of sets with two stillborn and the number expected under the independence hypothesis. In Figure 1a we present the data compiled by the authors, that is, the Swedish data and the data from England and Wales for the period 1996–2003.

We include in Figure 1b data for the Åland Islands, 1750 to 1949, Saxony, 1881 to 1900 (Lommatzsch, 1902), and for England and Wales, 1938 to 1948 (Lowe & Record, 1951). As pointed out above, our analyses of the data of Lowe and Record must be based on the additional assumption that the SBRs are the same for males and females. In both figures we use the SBR per 1000 as the argument.

We observe that the ratios between the observed and the expected numbers are markedly above the expected value of one, being almost always over three. When the SBR is low, the ratio is extremely high, especially for female–female sets. This indicates that when the SBR decreases, the number of sets with two stillborn decreases more slowly than expected according

Table 1

Observed Numbers of Male–Male Twin Pairs According to Live- and Stillbirths, in Sweden, 1869–1967

Period	Live–Live	Live–Still	Still–Still	Total		
Model	$n(1-p)^2$	$2np(1-p)$	np^2	n	$\hat{p}$	χ^2_1
1869–78	5103	888	264	6255	.1132	536.4
1901–10	5606	759	301	6666	.1021	957.1
1911–20	5156	686	258	6100	.0985	821.2
1921–30	4309	551	170	5030	.0886	519.9
1931–40	3380	454	138	3972	.0919	394.5
1941–50	4542	459	122	5123	.0686	458.0
1951–60	3641	288	64	3993	.0521	290.3
1961–67	2577	133	38	2748	.0380	314.8
Total	34,314	4218	1355	39,887	.0868	4430.0

Note: The probability $\hat{p}$ was estimated according to the independence model for stillbirths. The estimate is the relative frequency of stillbirths. Consequently, if it is multiplied by 1000 one obtains the SBR per 1000. The χ^2_1 values were obtained when the observed data were tested against the numbers estimated according to the model.

Table 2

Observed Numbers of Female–Female Twin Pairs According to Live- and Stillbirths, in Sweden, 1869–1967

Period	Live–Live	Live–Still	Still–Still	Total		
Model	$n(1-q)^2$	$2np(1-q)$	nq^2	n	$\hat{q}$	χ_1^2
1869–78	4962	676	194	5832	.0912	528.0
1901–10	5335	584	211	6130	.0821	828.3
1911–20	4874	518	169	5561	.0770	659.6
1921–30	4086	439	152	4677	.0794	600.0
1931–40	3317	394	115	3826	.0815	373.7
1941–50	4343	394	93	4830	.0600	371.4
1951–60	3322	208	46	3576	.0419	273.0
1961–67	2455	112	46	2613	.0390	480.2
Total	32,694	3325	1026	37,045	.0726	4113.7

Note: The probability $\hat{q}$ was estimated according to the independence model for stillbirths. The estimate is the relative frequency of stillbirths. Consequently, if it is multiplied by 1000 one obtains the SBR per 1000. The χ_1^2 values were obtained when the observed data were tested against the numbers estimated according to the model.

to the independence model. The points representing different countries in Figure 1a and 1b fit in well with the general pattern. Consequently, the dependence of the outcomes in the twin pairs causing an excess of twin sets containing two stillborns seems to be universal, following a certain trend.

In the Methods section we stressed that if it is possible to differentiate the SS twins into MZ and DZ twins, then the expected number of twin sets with two stillborn children is slightly increased. Preliminary studies of our different data sets indicate that the ratios are reduced by, at most, 20%. Such a reduction cannot solve the central problem concerning the high ratio values. Furthermore, if we assume that the independence holds within the MZ and DZ twin series, then the improvement of the goodness-of-fit is only minute and the deviations from the models are still statistically strongly significant. Furthermore, no similar reduction among OS twins is possible.

We also present an alternative attempt to consider the proportion of twin sets with two stillborn. Let this proportion be p_{22}. According to the independence model, $p_{22} = p^2$ among male–male twin sets, $p_{22} = q^2$ among female–female twin sets and $p_{22} = pq = \tilde{p}^2$ among OS twin sets. Consequently, the general formula should be $p_{22} = p^2$. In Figure 1a and 1b we observed an excess of twin sets with two stillborn. We tried to fit two alternative regression models, the first one, $p_{22} = \alpha + \beta p^2 + \varepsilon$, with an intercept and the second, $p_{22} = \beta p^2 + \varepsilon$, without the intercept. An excess of twin sets with two stillborn should result in an estimate $\hat{\beta} > 1$. We estimated the parameters in the models by weighted least squares. As weights the total number of twin sets was used. The argument for this is that the regressand p_{22} is a relative frequency and its variance is proportional to n^{-1} (Fellman & Eriksson, 1987). Figures 1a and 1b indicate that one can expect the same model to hold for different populations. Consequently, we considered all the data sets, but we

Table 3

Observed Numbers of Male–Female Twin Pairs According to Live- and Stillbirths, in Sweden, 1869–1967

Period	LM–LF	LM–SF	SM–LF	SM–SF	Total			
Model	$n(1-p)(1-q)$	$nq(1-p)$	$np(1-q)$	npq	n	$\hat{p}$	$\hat{q}$	χ_1^2
1869–78	6192	401	483	132	7208	.0853	.0739	194.3
1901–10	6912	305	349	146	7712	.0642	.0585	537.2
1911–20	6291	309	364	110	7074	.0670	.0592	272.4
1921–30	5216	242	296	103	5857	.0681	.0589	306.6
1931–40	4055	215	236	75	4581	.0679	.0633	178.0
1941–50	5099	205	259	45	5608	.0542	.0446	80.8
1951–60	4005	120	121	15	4261	.0319	.0317	28.3
1961–67	2629	49	59	17	2754	.0276	.0240	133.3
All	40,399	1846	2167	643	45,055	.0624	.0552	1730.1

Note: LM = live-born males, SM = stillborn males, LF = live-born females, SF = stillborn females.

The probabilities $\hat{p}$ and $\hat{q}$ were estimated according to the independence model for stillbirths. The estimates are the relative frequencies of stillbirths. Consequently, if they are multiplied by 1000, one obtains the SBRs per 1000. The χ_1^2 values were obtained when the observed data were tested against the numbers estimated according to the model.

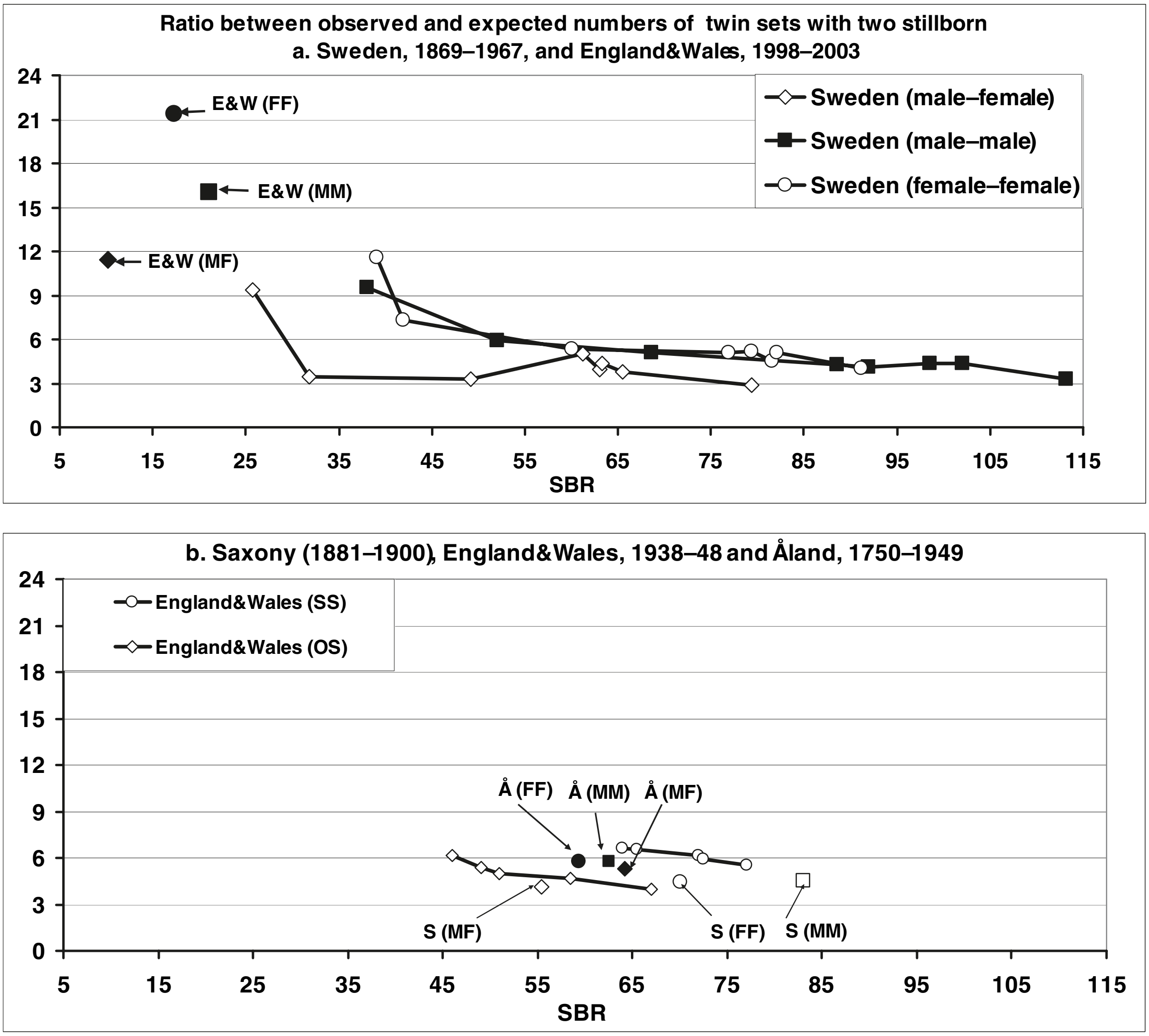

Figure 1

The ratio between the observed and expected number of twin pairs with two stillborn twins (MM = male–male; MF = male–female; FF = female–female; OS = opposite-sexed; SS = same-sexed)

Note: a. Data sets from Sweden, 1869–1967 and England and Wales (E&W) 1996–2003, both compiled for this study.

b. Data sets from the Åland Islands (Å), 1750–1949 (Eriksson, 1973); Saxony (S), 1881–1900 (Lommatzsch, 1902); and England and Wales, 1938–1948 (Lowe & Record, 1951). As pointed out in the text, Lowe and Record did not distinguish between males and females.

The stillbirth rate (SBR) as argument is given per 1000. The ratio increases with decreasing SBR. Consequently, the number of twin pairs with two stillborn decreases more slowly than expected when the SBR decreases. The points based on the Åland data and the data from England and Wales fit well within the general pattern.

analyzed SS, OS and all twin sets separately. The estimates are given in Table 4. We observe that, for both models, the differences between the β estimates for SS and OS data are not statistically significant. Consequently we used the whole material and estimated common parameters. Irrespective of the model chosen, estimate $\hat{\beta}$ is statistically significantly greater than one. This result supports our earlier finding that the proportion of twin sets with two stillborns differs from the expected. The intercept differs significantly from zero and, consequently, we have two alternative models, but both support our earlier conclusions. The model without an intercept seems to be more logical, because p_{22} has to be zero if $p = 0$. However, the positive intercept supports the observed increasing trend discussed in Figure 1a and 1b. In addition, the interval under study is ($10 \leq \text{SBR} \leq 113$) and, within this interval, the model with the intercept can also be considered relevant.

In Figure 2 we present the data and the curves based on both models and also include the curve $p_{22} = p^2$ corresponding to the independence model. All the data points observed are above that curve.

Triplet Sets

Above we have noted that if the probability of a stillborn male in a triplet set is p and of a stillborn female

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Table 4

Parameter Estimates of the Proposed Models

Model			$p_{22} = \beta p^2 + \varepsilon$	$p_{22} = \alpha + \beta p^2 + \varepsilon$	
	n		β	α	β
Same-sexed twin sets	27	Estimate	4.684	0.009165	3.349
	SE	0.209	0.001415	0.244	
	t	17.62	6.477	9.63	
Opposite-sexed twin sets	16	Estimate	4.065	0.002689	3.342
	SE	0.229	0.001287	0.403	
	t	13.38	2.090	5.81	
Total	43	Estimate	4.594	0.005181	3.745
	SE	0.164	0.001218	0.243	
	t	21.91	4.253	11.30	

Note: The *t* values were obtained when the intercept estimates were tested against zero and the regression parameter estimates against one.

is q, then the expected number of triplet sets with three stillborn children is $n(p + q)^3$. Among the 3594 triplets from Sweden, 1869 to 1967, 414 were stillborn. The estimated SBR for triplet males is $p = 0.1135$ and for females $q = 0.1163$. The observed number of triplet sets with three stillborn children is 34 and the estimated number is $n(p + q)^3 = 14.54$. Consequently, the ratio between the observed and the estimated number is 2.3. Among the 5670 triplets for England and Wales, 1996 to 2003, 157 were stillborn. The estimated SBR for triplet males is $p = 0.0301$ and for females $q = 0.0252$. The observed number of triplet sets with three stillborn children is six and the estimated number is $n(p + q)^3 = 0.319$. Consequently, the ratio between the observed and the estimated number is 18.8 (cf. Figure 1a, 1b). Lommatzsch (1902) presented triplets in Saxony, 1881 to 1900. He noted 345 triplet sets with known sex combination and known health condition and among these there were seven triplet sets with three stillborn. According to his data, we obtained an SBR among males of 0.1058 and among females 0.1284. Consequently, the expected number of sets with all stillborn is 4.43, and the ratio is 1.6.

Our studies of the twin and triplet data sets indicate that if the information concerning stillbirths among multiple births is only the relative frequency of

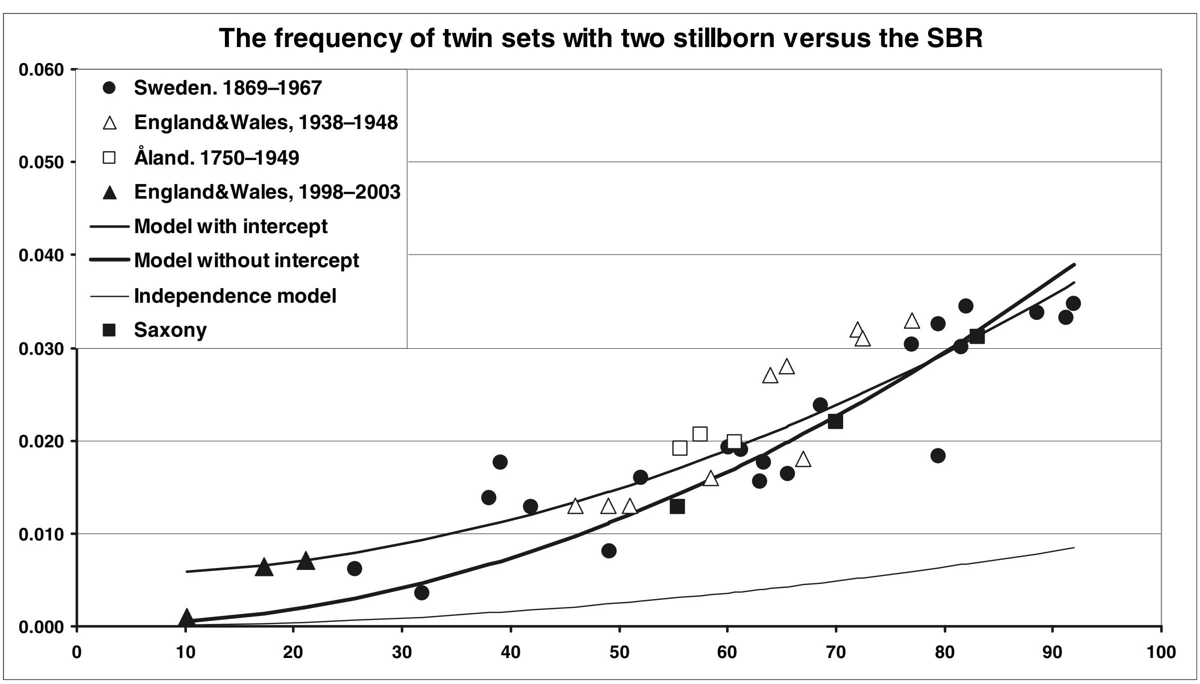

Figure 2

The probability of a twin set containing exclusively stillborn children versus the probability of a stillborn twin.

Note: The data sets are the same as in Figure 1.

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sets with two stillborns, then estimates of the SBRs based on this frequency have positive biases.

Discussion

Wedervang (1924, p. 259) noted that the proportion of twin maternities with two stillborn children is much higher among SS than among OS twin sets. Salihu et al. (2004) stated that siblings within a plurality set are generally subjected to similar prevailing conditions in the womb and are equally affected by the same attributes that characterize the mother. It is logical to expect conditions leading to the stillbirth of the member of a plurality set to have affected the other siblings as well, although unexplained factors determining vulnerability may vary. In short, one has to distinguish between the intracluster variation, that is the variation between individual twins or triplets, and the intercluster variation, that is the variation between mothers.

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