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# Effects of previous birth interval length on child outcomes can be estimated in a sibling analysis even when there are only two siblings 

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#### Abstract

Background: There is much interest in how the length of the previous birth interval affects various child outcomes, and it has become increasingly common to estimate such effects from sibling models. This is because one then controls for unobserved determinants of the outcome that are shared between the siblings and linked to the birth interval length. However, it is a common idea that such effects can only be estimated from data on families with three or more children. Objective: The goal of this paper is to show, through an intuitive argument and a simulation experiment, that it is possible to estimate effects of birth interval only from families with two children. Methods: Observations are simulated from two equations for fertility and one equation for child mortality. The fertility equations include a random term that is assumed to be correlated with the random term in the mortality equation. Mortality models are then estimated from the simulated observations. This is done 1000 times, and the averages of the 1000 sets of estimates are calculated. Results: The simulation experiment illustrates that it is indeed possible (by using a model specification that takes into account that no birth interval is defined for the first birth) to estimate birth interval effects in sibling models even when the data include only families with two children. Conclusion: It is good news that families with only two children can contribute to the estimation of birth interval effects. This is because, with a broader basis for the estimation, the precision is improved and there is less reason for concern about the general relevance of the estimates. An important limitation, however, is that it is potentially problematic to control for maternal age in a sibling model estimated only for the first and second child.


## KEYWORDS

birth interval, child outcome, sibling model

## 1 | BACKGROUND

There has been much interest in the importance of reproductive factors for child outcomes. For example, strong concerns have
been raised especially in developing countries about the health implications of being born to very young mothers or shortly after the previous birth, ${ }^{1,2}$ and the possible effects of high parental age on children's well-being have attracted increasing attention in low-fertility
settings. ${ }^{3}$ A key problem when analysing such effects is that several factors that are hard to measure affect both the mother's reproduction and the child outcomes. Many researchers have therefore estimated sibling models (also called sibling fixed effects, within-mother or within-family models), and thus controlled for unobserved parental and environmental characteristics that are constant over time and shared by the siblings. However, when this is done in investigations of how the length of the previous birth interval affects child outcomes, it is common to restrict the analysis to families with at least three children and leave out the first child..$^{4-10}$ The motivation is apparently that the previous birth interval is not defined for the first-born. This restriction of the estimation to families with at least three children is typically seen as a potential problem, both because it reduces the effective sample size and because one may suspect that the (social or biological) effects of the birth interval may not be the same for children of birth order three or higher, or for secondborn children with younger siblings, as they are for the second-born with no younger siblings.

However, if the standard assumption about constant unobserved confounders holds, it is indeed possible to estimate birth interval effects with sibling models even when the data include only families with two children, as was done in a recent Australian study ${ }^{11}$-although this study was immediately criticised. ${ }^{12}$ In short, the idea is that unobserved factors that both affect the reproductive process and have a constant impact on the child outcome-and that one controls for by doing a sibling analysis-not only add something to the outcomes for the second- and later-born children. They add the same to the outcome for the first-born. One can therefore take this source of confounding into account by comparing with the outcome for the first child, although one then, of course, has to use variables defined also for the first-born.

The possibility of including only the first and second child is explained in more detail below and illustrated through a simulation experiment. Infant mortality is the outcome in that simulation, because the importance of reproductive factors for infant and child mortality has attracted particularly much interest. However, when doing such a simulation, it is necessary to make the unrealistic assumption that fertility is not affected by earlier child deaths. This is because estimates from sibling models may actually be severely biased when a reproductive variable is the "exposure," and the "outcome" is mortality or something else that may affect subsequent fertility and thus the "exposure" for a younger sibling-a methodological problem that has only recently been acknowledged in the literature. ${ }^{13}$ This has, of course, no implications for the conclusions that are drawn. The same picture would have appeared from the simulations if another outcome not affecting the "exposure" for younger siblings in the real world had been chosen.

In the simulations, constant unobserved determinants of infant mortality are assumed to be linked to the reproductive factors, which is a structure that would motivate a sibling analysis. More specifically, the simulation model includes equations for first and higher-order birth probabilities (which we may consider as generating the reproductive variables) and an equation for infant mortality.

## Synopsis

## Study question

Is it possible to estimate effects of previous birth interval length on child outcomes in sibling models when there are only two siblings?

## What's already known

It has become increasingly common to use sibling models when analysing effects of birth interval length and other reproductive factors on child outcomes, because one then controls for unobserved constant family characteristics. However, it is a common idea that only families with three or more children can contribute when estimating such effects.

## What this study adds

It is explained, and illustrated through simulation experiments, that it is possible to estimate birth interval effects in sibling models even when the data include only families with two children, although control for maternal age in such models is potentially problematic.

The equations include random terms that represent time-invariant unobserved characteristics of the mother and her environment, and that are allowed to be correlated.

In a final step, models for preterm birth (a more common outcome in rich countries) are estimated from real data-for women with two children and for women with three or more children. However, although the issue is important and findings from earlier sibling analyses have been mixed, ${ }^{5-9}$ the results will be commented on only from a methodological perspective.

## 2 | METHODS

## 2.1 | An intuitive explanation

Consider a continuous outcome $y_{i j}$ for child $i$ in family $j$ given by.

$$
\begin{equation*}
y_{i j}=\lambda_{0}+\lambda C_{i j}+e_{i j} \tag{1}
\end{equation*}
$$

where $e_{i j}$ is a normally distributed random term with zero mean and $C_{i j}$ is a categorical variable representing a combination of birth order, which is 1 or 2 , and birth interval length, which is short, medium, or long. (Categorical variables, which can be considered as vectors of $0 / 1$ dummies, and the associated coefficients are symbolised with bold types.) A first-born child is in the first category of $C_{i j}$, a second child born after a short interval is in the second category, a second
child born after a medium interval is in the third category, and a second child born after a long interval is in the fourth category. If the first category is chosen as the reference category, the four coefficients (or "effects") that constitute $\lambda$ are $0, \lambda_{1}, \lambda_{2}$, and $\lambda_{3}$. In other words, if we set the error term to its average value of 0 (this condition is ignored below for simplicity), the outcome for the first child is $\lambda_{0}$ and that for the second child is $\lambda_{0}+\lambda_{1}, \lambda_{0}+\lambda_{2}$, or $\lambda_{0}+\lambda_{3}$, depending on whether the interval is short, medium, or long. The difference in the outcome between a second child born after a short interval and a second child born after a medium interval, which is $\lambda_{1}-\lambda_{2}$, can be referred to as the effect of being born after a short rather than a medium interval, given birth order (in this case two). Similarly, $\lambda_{3}-\lambda_{2}$ is the effect of a long versus medium interval.

This situation-with the outcomes for the four types of children being $\lambda_{0}, \lambda_{0}+\lambda_{1}, \lambda_{0}+\lambda_{2}$, and $\lambda_{0}+\lambda_{3}$-can also be described by the model

$$
\begin{equation*}
y_{i j}=\mu_{0}+\mu_{1} S_{i j}+\mu_{2} D_{i j}^{\prime}+e_{i j} \tag{2}
\end{equation*}
$$

$S_{i j}$ is a dummy that is 1 if the child is second-born and 0 if the child is first-born. $D^{\prime}{ }_{i j}$ is a birth interval variable with the three categories short, medium, and long, where medium arbitrarily is chosen as the reference category, and a first-born child is put in the reference category. $\mu_{0}=\lambda_{0}, \mu_{1}=\lambda_{2}$, and $\mu_{2}$ (which is a vector) is $\lambda_{1}-\lambda_{2}$ for the short-interval category, 0 for the medium-interval category and $\lambda_{3}-\lambda_{2}$ for the long-interval category. The model can, of course, be generalised to families with more than two children. One can then define $S_{i j}$ as having the two categories 1 and 2 or more, but it would be even more reasonable to use a birth order variable with more than two categories (such as $\mathbf{O}_{i j}$ in the simulation below), because long intervals tend to be more frequent at higher birth orders, and one will typically want to control for that.

Model (2) can alternatively be written on interaction form as

$$
\begin{equation*}
y_{i j}=\zeta_{0}+\zeta_{1} S_{i j}+\zeta_{2} S_{i j} D_{i j}+e_{i j} \tag{3}
\end{equation*}
$$

where $D_{i j}$ needs not be defined for the first-born (for whom $S_{i j}=0$ ) because the term then cancels out (which is the reason for writing $\boldsymbol{D}_{i j}$ instead of $\boldsymbol{D}^{\prime}{ }_{i j}$ as above). The outcomes for the four types of children will be as above if $\zeta_{0}=\mu_{0}=\lambda_{0}, \zeta_{1}=\mu_{1}=\lambda_{2}$, and $\zeta_{2}$, which is a vector, is $\lambda_{1}-\lambda_{2}$ for the short-interval category, 0 for the mediuminterval (reference) category and $\lambda_{3}-\lambda_{2}$ for the long-interval category. This was essentially the parameterisation that was used in the Australian study. ${ }^{11}$

Let us now, for simplicity, make the additional assumption that there is "something unobserved" with all mothers who have a short birth interval that also increases the outcome by $\delta$ for all their children including the first. Thus, if we again set the error term to 0 , the outcome for these mothers' first child is $\lambda_{0}+\delta$ and the outcome for their second child is $\lambda_{0}+\lambda_{1}+\delta$. In contrast, let us assume that there is no such additional contribution for mothers who have a medium interval between the births, so the outcomes for their two children are $\lambda_{0}$ and $\lambda_{0}+\lambda_{2}$. The motive for doing sibling analysis is to handle
this kind of situation, where the difference between the outcome for a second child born after a short interval and that for a second child born after a medium interval-which would be the effect estimated from a model such as (2) without sibling fixed effects-is $\lambda_{0}+\lambda_{1}+\delta$ $-\left(\lambda_{0}+\lambda_{2}\right)=\lambda_{1}+\delta-\lambda_{2}$, while the true effect still is $\lambda_{1}-\lambda_{2}$. When a sibling fixed effects version of (2) is estimated, one may say that a correct estimate of $\mu_{1}=\lambda_{2}$ "comes from" comparing among mothers who have a medium birth interval; it is the difference in the outcome between their first and second child. The effect of being born after a short rather than medium interval (ie the first element of $\mu_{2}$ ) is correctly estimated by taking the difference between $\lambda_{1}$, which is the difference in the outcome between the first and second child among women with a short interval, and $\lambda_{2}$. A similar argument can be made for the estimation of the effect of having long rather than medium interval.

Hutcheon and Harper ${ }^{12}$ made two points in their criticism of the Australian study. First (when their argument is transformed to fit with the example above), they stated that the difference between $\lambda_{1}$ and $\lambda_{2}$ is not the effect of having short versus medium interval, but the difference in the effect of birth order (two vs one) between those with short and those with medium interval. However, both interpretations are reasonable. As mentioned above, the difference $\lambda_{1}-\lambda_{2}$ in the outcome between second-born children with short and medium interval-in a "world" as described by the models (1), (2), or (3)-is precisely what one usually means by an effect of short vs medium interval, given birth order (in this case two). In a "world" where there is a contribution $\delta$ from an unobserved confounder such as described above, it would still be reasonable to consider $\lambda_{1}-\lambda_{2}$ as the (true) birth interval effect, and it is this effect that appears when a sibling fixed effect version of models such as (2) and (3) are estimated.

Their second main point was that the birth interval effect that is estimated with a sibling model is not the true effect (ie the constant unobserved confounders are not taken adequately into account), because the estimate is based on comparisons across mothers. However, sibling model estimation means that both $\lambda_{1}$ and $\lambda_{2}$ are correctly estimated from within-mother comparisons (although from different mothers), and the difference between the estimates is therefore also an unbiased within-mother estimate of $\lambda_{1}-\lambda_{2}$, which is the effect of short versus medium birth interval.

To be convinced that a sibling analysis indeed will work when there are only two siblings, one may set up a population with very simple features such as just mentioned, or alternatively carry out a simulation that reflects a more realistic process. Such a simulation, which involves infant mortality as the outcome, is presented below.

## 2.2 | Simulation

The "data generating model" included the Equations 4-6 below. More specifically, each simulation started with 100000 women just turned 17 years old. For each woman (j), fertility and mortality random termsassumed to affect her fertility throughout her reproductive period and
TABLE 1 Effects of birth interval and birth order on infant mortality

| Variables | Effects in simulation | Average of estimates from sibling models |  |  |  |  |
| :---: | :---: | :---: | :---: | :---: | :---: | :---: |
|  |  | Mothers with three or more children, but first child excluded | Mothers with three or more children | Mothers with two children | All mothers | All mothers, but only first and second child included |
| Birth interval (mo) |  |  |  |  |  |  |
| 1-18 | 0.57 | 0.58 | 0.58 | 0.56 | 0.58 | 0.57 |
| 19-27 | 0.20 | 0.21 | 0.21 | 0.22 | 0.21 | 0.21 |
| 28-36 ${ }^{\text {a }}$ | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) |
| 37-48 | -0.20 | -0.19 | -0.19 | -0.20 | -0.19 | -0.20 |
| 49-60 | -0.14 | -0.13 | -0.14 | -0.15 | -0.14 | -0.14 |
| 61-72 | -0.10 | -0.10 | -0.09 | -0.12 | -0.09 | -0.10 |
| 73-84 | -0.08 | -0.08 | -0.08 | -0.06 | -0.08 | -0.08 |
| 85-96 | -0.08 | -0.07 | -0.06 | -0.10 | -0.07 | -0.09 |
| 97-108 | -0.08 | -0.08 | -0.07 | -0.07 | -0.07 | -0.08 |
| 109-120 | -0.08 | -0.08 | -0.08 | -0.05 | -0.08 | -0.08 |
| 121- | -0.08 | -0.08 | -0.08 | -0.08 | -0.08 | -0.09 |
| Mean absolute bias ${ }^{\text {b }}$ |  | 0.005 | 0.007 | 0.014 | 0.006 | 0.003 |
| Birth order |  |  |  |  |  |  |
| 1 | 0.00 (Reference) |  | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) |
| 2 | -0.11 | $\begin{aligned} & 0.00 \\ & \text { (Reference) } \end{aligned}$ | -0.12 | -0.12 | -0.12 | -0.12 |
| 3 | 0.14 | 0.25 | 0.14 |  | 0.14 |  |
| 4 | 0.50 | 0.61 | 0.50 |  | 0.50 |  |
| 5 | 0.50 | 0.61 | 0.50 |  | 0.50 |  |

${ }^{\text {a }}$ The reference category for birth interval includes first-born children.
${ }^{\text {b }}$ The "bias" refers the difference between an effect parameter used in the simulation and the average of the corresponding estimates, and the mean is taken over all the 10 parameters for birth interval.
TABLE 2 Effects of birth interval and birth order on infant mortality

| Variables | Effects in simulation | Average of estimates from naive models |  |  |  |  |
| :---: | :---: | :---: | :---: | :---: | :---: | :---: |
|  |  | Mothers with three or more children, but first child excluded | Mothers with three or more children | Mothers with two children | All mothers | All mothers, but only first and second child included |
| Birth interval (mo) |  |  |  |  |  |  |
| 1-18 | 0.57 | 0.73 | 0.73 | 0.64 | 0.76 | 0.87 |
| 19-27 | 0.20 | 0.30 | 0.30 | 0.26 | 0.32 | 0.38 |
| 28-36 ${ }^{\text {a }}$ | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) |
| 37-48 | -0.20 | -0.27 | -0.27 | -0.25 | -0.28 | -0.34 |
| 49-60 | -0.14 | -0.29 | -0.29 | -0.26 | -0.31 | -0.42 |
| 61-72 | -0.10 | -0.28 | -0.28 | -0.25 | -0.31 | -0.45 |
| 73-84 | -0.08 | -0.30 | -0.30 | -0.25 | -0.33 | -0.49 |
| 85-96 | -0.08 | -0.30 | -0.30 | -0.29 | -0.33 | -0.54 |
| 97-108 | -0.08 | -0.31 | -0.31 | -0.27 | -0.35 | -0.55 |
| 109-120 | -0.08 | -0.32 | -0.32 | -0.28 | -0.36 | -0.58 |
| 121- | -0.08 | -0.30 | -0.30 | -0.27 | -0.36 | -0.58 |
| Mean absolute bias ${ }^{\text {b }}$ |  | 0.179 | 0.179 | 0.141 | 0.210 | 0.359 |
| Birth order |  |  |  |  |  |  |
| 1 | 0.00 (Reference) |  | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) | 0.00 (Reference) |
| 2 | -0.11 | $\begin{aligned} & 0.00 \\ & \text { (Reference) } \end{aligned}$ | -0.14 | -0.07 | 0.01 | -0.01 |
| 3 | 0.14 | 0.29 | 0.15 |  | 0.49 |  |
| 4 | 0.50 | 0.85 | 0.71 |  | 1.05 |  |
| 5 | 0.50 | 0.99 | 0.85 |  | 1.18 |  |

${ }^{\text {a }}$ The reference category for birth interval includes first-born children.
"The "bias" refers the difference between an effect parameter used in the simulation and the average of the corresponding estimates, and the mean is taken over all the 10 parameters for birth interval.
all her children's infant mortality-were drawn from a bivariate normal distribution with zero mean. Both variances were set to 1 and the correlation to 0.75 , but the choice of variances and correlation is not important for the conclusion. In other words, there is a fertility random term reflecting constant unobserved determinants of fertility and a mortality random term reflecting constant unobserved determinants of mortality, and the two are correlated to reflect that some of these constant unobserved determinants affect both fertility and mortality. At each month ( $t$ ) until the woman turned 45 , probabilities of first $\left(f_{1 t j}\right)$ and higher-order ( $f_{2 t j}$ ) births were predicted from the logistic equations

$$
\begin{gather*}
\log \left(f_{1 \mathrm{tj}} /\left(1-f_{1 t \mathrm{j}}\right)\right)=\alpha_{0}+\boldsymbol{\alpha}_{1} \mathbf{A}_{t j}+\sigma_{j}  \tag{4}\\
\log \left(f_{2 \mathrm{tj}} /\left(1-f_{2 t j}\right)\right)=\beta_{0}+\boldsymbol{\beta}_{1} \mathbf{A}_{t j}+\boldsymbol{\beta}_{2} \boldsymbol{D}_{t j}+\beta_{3} \mathbf{P}_{t j}+\sigma_{j} \tag{5}
\end{gather*}
$$

where $\boldsymbol{A}_{\mathrm{tj}} \boldsymbol{D}_{\mathrm{tj}}$ and $\boldsymbol{P}_{\mathrm{tj}}$ are categorical variables representing age, time since last birth, and parity, respectively, and $\sigma_{j}$ is the fertility random term. The coefficients (ie the $\alpha \mathrm{s}$ and $\beta \mathrm{s}$ ) were taken from an earlier study ${ }^{13}$ and reflect patterns actually observed in Norway, but subjected to visual smoothing. The variables $\boldsymbol{A}_{\mathrm{t} j} ; \boldsymbol{D}_{\mathrm{t} j}$, and $\boldsymbol{P}_{\mathrm{t} j}$ were updated monthly as the simulation "proceeded."

For each month, a number was drawn from a uniform distribution over [ 0,1$]$, and if this number was smaller than the relevant predicted birth probability, a birth (up to a fifth, as few have more children in Norway) was assigned to the woman that month. In that case, the probability that this child died within 12 months was predicted from the relevant demographic variables multiplied by coefficients taken from the mentioned earlier study ${ }^{13}$ (except that a larger intercept was used to get a larger number of deaths), and with a mortality random term added-in parallel with the predictions of fertility. In mathematical terms, the prediction equation for the death probability $m_{i j}$ for child $i$ of mother $j$ was:

$$
\begin{equation*}
\log \left(m_{i j} /\left(1-m_{i j}\right)\right)=\gamma_{0}+\gamma_{1} D_{i j}^{\prime}+\gamma_{2} O_{i j}+\tau_{j} \tag{6}
\end{equation*}
$$

where $D_{i j}{ }^{\prime}$ is time between current and previous birth (primed because the reference category includes the first-born child), $O_{i j}$ is birth order (ie the mother's parity $(\boldsymbol{P})$ the month after the child was born), and $\tau_{j}$ is the mortality random term. Again, a number was drawn from a uniform distribution over [0,1], and if this was smaller than the predicted death probability, a death was assigned to the child.

Note that this mortality equation is just as (2) in the discussion above, except that birth order has more categories than 1 and 2 and a random term has been added.

## 2.3 | Estimation of mortality models from the simulated population

Logistic mortality models were estimated from the simulated population. The equation was as above, except that the random term was substituted by sibling fixed effects $\left(\nu_{j}\right)$ :

$$
\begin{equation*}
\log \left(m_{i j} /\left(1-m_{i j}\right)\right)=\eta_{0}+\eta_{1} \boldsymbol{D}_{i j}^{\prime}+\eta_{2} \boldsymbol{O}_{i j}+v_{j} \tag{7}
\end{equation*}
$$

The estimation was done with Proc Logistic in SAS, using the Strata command for conditional likelihood maximisation. For comparison, also some models without the sibling fixed effects (referred to below as "naïve models") were estimated.

Simulation followed by estimation was done 1000 times, after which the averages over the 1000 sets of estimates were calculated. These averages are shown in the tables along with the mortality effect coefficients used in the simulation and the mean absolute bias.

## 2.4 | Estimation from real data

Models such as (7) were estimated for preterm births, using data on live births between 1967 and 2015 from the Norwegian Medical Birth Register. Two groups of women were included: (a) those who had their first and second child (but no higher-order births) with the same father during the 1967-2015 period, and (b) those who had three or more children with the same father within this period, although the first child was not included in the analysis. A focus on interpregnancy rather than interbirth interval was possible with these data. The use of data for this purpose has been approved by the Regional Committees for Medical and Health Research Ethics and the data owners.

## 3 | RESULTS

## 3.1 | Results from the simulation experiment

If only mothers with three or more children were included in the analysis, the sibling model gave estimates close to the true effects (Table 1, column 1)-regardless of whether the first-born was included (column 3), as some people might assume should be avoided, or left out (column 2). The estimates were also correct if only mothers with two children were included (column 4).

Additionally, sibling models were estimated from a larger sample by including both mothers with two children-who we have now seen can contribute in the estimation-and those with three or more. As one would expect, these estimates (column 5) were also very close to the true effects, as were those obtained when only the first two children in all these families were included (column 6).

The corresponding naïve models (ie without sibling fixed effects) were severely biased (Table 2).

The shape of the associations between birth interval and the various outcomes that have been analysed in earlier studies will typically be captured best by a categorical specification. However, in a supplementary simulation experiment, the birth interval was arbitrarily set to 32 months for the first-born, and the term -0.02 (interval-32) +0.0001 (interval-32) ${ }^{2}$ was included in the simulation equation instead of $\gamma_{1} D^{\prime}$. This second-degree polynomial mimics the shape of the birth interval effect in $\gamma_{1}$ quite well. The estimated
coefficients were very close to the true ones even when only mothers with two children were included (-0.0199 and 0.000100; not shown in tables).

In a final step, it was experimented with effect modification: the effect of birth interval assumed so far to be general was now restricted to the second-born children, while a more sharply negative effect was assumed for third- and later-born. When a corresponding interaction between birth order and birth interval was added to a model estimated from mothers with three or more children, this interaction and the main effect were, as expected, almost exactly as assumed in the simulation (not shown in tables)regardless of whether the first-born child was included. When a model was estimated only for those with two children, the main effect was correctly estimated, while it was not relevant to include an interaction.

## 3.2 | Results from analysis of real data

For both groups of women, the naïve model suggested a relatively high risk of preterm birth for children born after a very short interval, plus a rising risk as the interval increases beyond three years (Table 3). When a sibling model was estimated instead, the excess short-interval risk was strongly reduced for both groups. Also, the
higher risk at longer intervals was less pronounced, most clearly among two-child mothers.

## 4 | COMMENT

## 4.1 | Principal findings

It was argued theoretically that effects of birth interval can be correctly estimated from information on only the first and second child when the unobserved factors linked to reproduction affect the outcomes for first- and later-born children similarly. The simulation experiment (with birth order and birth interval as the only observed determinants) supports that idea, and in an example based on real data the implications of adding sibling fixed effects did not differ very much depending on whether the analysis included two-child mothers or women with more children.

## 4.2 | Strengths and limitations

It is valuable to see the theoretical arguments so clearly backed up by large-scale simulation experiments based on realistic effect coefficients and that estimation from high-quality register data points

TABLE 3 Effects (odds ratios with $95 \%$ confidence interval) of birth interval and birth order on the chance of preterm birth, according to data from the Norwegian Medical Birth Register, 1967-2015

| Variables | Estimates from sibling models |  | Estimates from naïve models |  |
| :---: | :---: | :---: | :---: | :---: |
|  | Mothers with three or more children, but first child excluded | Mothers with two children | Mothers with three or more children, but first child excluded | Mothers with two children |
| Interpregnancy interval (mo) |  |  |  |  |
| 1-9 | 1.14 (1.06, 1.23) | 1.08 (1.00, 1.18) | 1.48 (1.41, 1.56) | 1.43 (1.35, 1.52) |
| 10-18 | 1.03 (0.96, 1.10) | 1.03 (0.96, 1.10) | 1.05 (1.00, 1.10) | 1.04 (0.99, 1.09) |
| $19-27^{\text {a }}$ | 1.00 (Reference) | 1.00 (Reference) | 1.00 (Reference) | 1.00 (Reference) |
| 28-36 | 0.95 (0.88, 1.03) | 0.98 (0.92, 1.05) | 1.02 (0.97, 1.08) | 1.08 (1.03, 1.13) |
| 37-48 | 1.08 (1.00, 1.16) | 1.10 (1.03, 1.18) | 1.12 (1.06, 1.18) | 1.20 (1.51, 1.26) |
| 49-60 | 1.14 (1.05, 1.25) | 1.21 (1.11, 1.32) | 1.23 (1.16, 1.31) | 1.38 (1.30, 1.47) |
| 61-72 | 1.41 (1.26, 1.58) | 1.38 (1.24, 1.53) | 1.35 (1.26, 1.45) | 1.56 (1.46, 1.68) |
| 73-84 | 1.51 (1.32, 1.71) | 1.49 (1.30, 1.70) | 1.45 (1.34, 1.58) | 1.68 (1.54, 1.83) |
| 85-96 | 1.80 (1.54, 2.10) | 1.77 (1.50, 2.09) | 1.71 (1.55, 1.88) | 2.01 (1.80, 2.23) |
| 97-108 | 1.80 (1.51, 2.16) | 1.79 (1.45, 2.20) | 1.83 (1.64, 2.05) | 2.19 (1.92, 2.50) |
| 109-120 | 1.54 (1.23, 1.93) | 1.99 (1.54, 2.55) | 1.75 (1.52, 2.02) | 2.47 (2.11, 2.89) |
| 121- | 2.02 (1.70, 2.40) | 2.51 (2.06, 3.07) | 2.26 (2.03, 2.51) | 2.73 (2.42, 3.08) |
| Birth order |  |  |  |  |
| 1 |  | 1.00 (Reference) |  | 1.00 (Reference) |
| 2 | 1.00 (Reference) | 0.62 (0.60, 0.65) | 1.00 (Reference) | 0.61 (0.59, 0.64) |
| 3 | 1.01 (0.97, 1.04) |  | 1.01 (0.98, 1.04) |  |
| 4 | 1.23 (1.14, 1.31) |  | 1.27 (1.20, 1.34) |  |
| 5 | 1.44 (1.24, 1.67) |  | 1.43 (1.27, 1.61) |  |

${ }^{\text {a }}$ The reference category for interpregnancy interval includes first-born children.
in the same direction. However, it has not been illustrated through simulations how biased the estimates might be in various situations where the unobserved confounders are not constant (ie shared by all siblings). More importantly, maternal age has been left out of the simulation and estimation even though it is often taken into account in studies of birth interval effects. The reason is that there is a linear dependence between variables when maternal age is added to sibling models estimated for only the first and second child (but not when the model is estimated for larger families): Mother's age minus the birth interval variable multiplied with a dummy for second birth is the mother's age when the first child was born, which is the same for both siblings and can be seen as part of the sibling fixed effect. This means that effects of maternal age and birth interval may be hard to separate. The bias may in some cases be large and in other cases of no practical importance, depending on how the effects are in reality and the specifications of the model that is estimated. This is a complicated issue that needs exploration in future studies. See details in the Appendix S1. (Note also that a similar linear dependence problem makes it problematic to separate effects of maternal age and birth year in sibling models, and not only when the analysis is restricted to the first two children ${ }^{14}$ ).

## 4.3 | Interpretations

It is a common idea that only families with three or more children can be included (and the first child left out) when birth interval effects are estimated from sibling models. ${ }^{5-10}$ However, by using model specifications that take into account that a birth interval is not defined for the first-born, it is indeed possible to estimate interval effects for the second child from families with only two children. This is good news because even in Norway, where fertility is higher than in most other rich countries, only $29 \%$ of the women who are now 45 years old have had three or more children, and only $49 \%$ of the children born to mothers in this birth cohort would be included in an analysis restricted to mothers with more than two children (and $30 \%$ of these children would be first-born). ${ }^{15}$ Adding the large group of women with two children would make the estimates more precise. Furthermore, it would no longer be relevant to claim that the estimates reflect only the situation in families with at least three children; they reflect the combination of that situation and that in families with only two children.

However, if an analysis is based only on the first and second child, one can, of course, only learn about the effect of the interval preceding the second birth. It is possible that interval effects vary with birth order, and to explore such effect modification one has to include also later-born children and add interaction terms.

One should also keep in mind that sibling analysis rests on an assumption about unobserved factors having the same effects on all siblings. If there, in reality, are time-varying factors of importance for the birth interval length that also affect the child outcome, and these are not controlled for, the sibling model estimates will no longer be unbiased. A sibling analysis of the first and second child necessarily includes a main effect of birth order. If there is no substantive interest
in the birth order effect, it is not necessary to control for time-varying factors that affect the chance of having another child, and not the birth interval, but one can hardly ever be sure about the latter. Thus, it would be a good strategy to control for all time-varying fertility determinants to the extent that it is possible. It should be noted, however, that the same applies to analysis that does not include the first child. Birth order is likely to be (positively) correlated with birth interval length, so unless it can be assumed to have no impact on the outcome, one should control for it. Also, there are the same arguments for controlling for time-varying fertility determinants.

It would make good sense and is indeed also very common, to control for maternal age when estimating birth interval effects. Such control would be particularly important if the interest lies in the effects of very long intervals, since women who had their previous birth several years earlier obviously cannot be very young. Very short intervals are less clearly linked to maternal age, although they may be more common at a low age, even when comparison is made within mothers. However, one should not control for maternal age in a sibling analysis of birth interval effects that is restricted to the first and second child-unless it is shown in future studies that the bias because of the linear dependence between maternal age, the birth interval variable, and the sibling fixed effect is small in the relevant situation. Stated differently, if only data about the first and second child are available, the choice (according to current knowledge) is between a sibling model without control for maternal age or a "naïve" model-with its obvious disadavantages-where such control is included. The former may well be seen as the best alternative. If mothers who have more children, for whom there is no such linear dependency problem, are included in a sibling analysis along with two-child mothers it may be quite acceptable to control for maternal age, but this remains to be checked properly.

## 5 | CONCLUSION

Researchers analysing the impact of birth interval length on child outcomes often compare second- and later-born children, but if the standard assumption about constant unobserved confounders holds, one can also analyse two-child families. It would be valuable to check in future empirical studies whether estimates based on the first and second child indeed tend to be quite similar to those obtained from larger families. Also, it is important to identify through simulations whether there are situations where it is acceptable to control for maternal age when estimating models for two-child families.

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## CONFLICT OF INTERESTS

None declared.

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## SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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