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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 15,611 Amish couples born during 1725-1925 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure Hazard ratios of couples with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both widows and widowers. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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3 socially cohesive Amish community. Remarriage is associated with a significant
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5 decrease in the mortality risk among widowed individuals. Contrary to results
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8 from previous studies, an increase in the number of surviving children was
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10 associated with decreased survival rate.
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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.
- We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks of the bereaved individuals and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of a large and uncensored dataset, our hazard ratio estimates are mostly significant with narrow confidence intervals.
- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.

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- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.

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INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.^{2 5} Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6 7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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To gain a different perspective on potential influences of social support on

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3 bereavement, we assessed the bereavement effect in an Amish population, whose
4 culture is characterized by its core beliefs in community and social cohesion.
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6 Examples of the strong degree of social support within the Amish include
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8 community-managed health insurance and community ties through membership in
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10 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
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12 We reasoned that the high level of social support in the Amish population might
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14 mitigate the bereavement effect. Previous studies used covariates such as
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16 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
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18 neighborhood interaction, and household size to study the relationship between
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20 bereavement and social support.^{4 8 13}
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27 We estimated bereavement effects in widows and widowers separately, and
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29 further assessed mortality risks as a function of age at widowhood and time since
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31 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
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33 association of bereavement and mortality of widowed husbands and wives. We
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35 considered remarriage and number of surviving children as additional potential
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37 modifiers of the bereavement effect.
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43 MATERIAL AND METHODS

44 Data source

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46 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
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48 effect of bereavement on the Amish population.^{14 15} The AGDB is a
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50 computerized genealogy database of Amish and some Mennonite populations, last
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3 updated in 2010. The AGDB contains information for 539,822 Amish individuals
4 and 136,213 Amish couples. This database includes information about the family
5 relationships and the birth, marriage, and death dates of children of Amish
6 individuals from North America, mostly located in Pennsylvania, Ohio, and
7 Indiana.
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15 Amish people rarely give birth out of wedlock, and a vast majority of the
16 mating samples we used are documented as marriages in the three original sources
17 from which AGDB is derived. However, there is less availability of marriage
18 dates than birth dates; we did not wish to exclude couples with missing marriage
19 dates. For this study, we included couples in which both spouses met four
20 conditions: (1) they were born prior to January 1, 1926; (2) their birth and
21 death dates were recorded; (3) they were in their first marriage; and (4) they did
22 not die on the same day. Constraint (4) excludes couples who may have died from
23 a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion
24 criteria. The oldest person included was born on February 14, 1725, and the
25 youngest on December 31, 1925.
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41 Our data set included 15,611 couples with information on their date of birth,
42 date of death, number of surviving children, family IDs, and remarriage status
43 after widowhood. For some analyses, these couples were first partitioned into
44 eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-
45 1900, and 1901-1925) and sex. For the time since bereavement analyses, we did
46 not partition into cohorts because the number of widow(er)s who died within 6
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3 months after being bereaved is small (N=224). Characteristics of these couples by
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5 husband birth cohort are shown in Table 1.
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8 9 **Statistical analyses**

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11 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
12 Hazards (CPH) model to study the association of widowhood and mortality rates
13 in the surviving spouse, while adjusting for covariates such as education, health
14 habits, age in years, number of children, and remarriage. In some of our analyses,
15 we adjusted for remarriage and number of children as covariates.
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23 The main covariate, widowhood, was monitored as a time-dependent
24 covariate, by assigning a value of 1 for each time period the individual was
25 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
26 covariate, by assigning a value 1 for the widowed period if the individual
27 remarried and 0 otherwise. The number of surviving children was a time-
28 independent covariate and divided into categories, as follows: 0=number of
29 surviving children is ≤ 2 , 1=number of surviving children is between 3 and 6, and
30 2=number of surviving children is > 6 .
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42 Survival analyses were performed using the R programming environment.¹⁷
43 All estimates and confidence intervals were obtained using the `coxph` function
44 available in the `survival` package.
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49 **Representation of survival data for CPH analysis**

50 We show below examples of how we represented the survival data for CPH
51 analysis using a time modeling approach. To represent the survival data columns, the
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standard approach is to convert the couple's demographic information, date of death, date of birth, remarriage and number of surviving children into columns representing **start** time, **stop** time, **event** status, and all of the included covariates (widowhood, remarriage and number of surviving children).^{18 19} Here (**start**, **stop**] is an interval of risk, open on the left and closed on the right; the **event** column is set to 1 if the subject had an event (death) at time **stop** and is 0 otherwise. To illustrate the subtleties, we consider a hypothetical example of three wives (ID1, ID2 and ID3) from the cohort 1850-1875.

- *ID*1: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried; number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
- *ID*2: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
- *ID*3: born on 03/01/1871; never widowed and never remarried; number of surviving children = 4 and died on 04/01/1930 at age 59.

The data used for the CPH considering widowhood as a single time-dependent covariate is presented in Table 2a. The following model was used to estimate the association between widowhood and mortality.

$$h(t) = h_0(t) \exp[\beta W]$$

This model can be run with additional covariates and results that adjust for the ability to remarry and the number of surviving children are presented.

Timing of widowhood

The above model considers widowhood as a single time-dependent covariate, evaluating the association between mortality and being widowed. One can further ask if that association varies over the life course by considering the impact of age at widowhood on mortality. We addressed this issue following the approach of Mineau et al. by adding terms for age at widowhood into the model as covariates (<45, 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to age at widowhood (see Table 2b).⁸ These terms were included in the model as time-dependent dummy variables, created to represent the widowhood experience of each individual across the 5 age windows spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} . Further, we also addressed the impact of remarriage and number of surviving children on mortality of widowed individuals.

The following model can be used to estimate the association between each of the five dummy variables and mortality:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , W_{75+} columns provided in the Table 2b.

Time since bereavement

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Next, we evaluated the association between mortality and time since bereavement or widowhood. We followed the approach of Schaefer et al. by considering the following time since bereavement ranges: 0-6 months, 7-12 months, 13-24 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This approach allowed us to estimate HR according to time since bereavement (see Table 2c). The columns TSB₀₋₆, TSB₇₋₁₂, ... in Table 2c are time dependent covariates that change with the survival time associated with widowed husbands and wives. We did not account for remarriage in this analysis because of missing remarriage dates.

The following model was used to study the association between mortality and time since bereavement:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

where W_1, \dots, W_7 dummy variables are associated with the TSB₀₋₆, TSB₇₋₁₂, TSB₁₃₋₂₄, TSB₂₅₋₃₆, TSB₃₇₋₄₈, TSB₄₉₋₆₀ and TSB_{>60} columns provided in the Table 2c.

RESULTS

We initially partitioned the 15,611 couples from AGDB into four cohorts ranging in size from 3,210 (husband birth year 1850-1875) to 4,466 (husband birth year 1876-1900). Wives were more likely to die after their husbands in all cohorts, although in the three earliest cohorts (prior to 1850, 1850-1875, and

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3 1876-1900), husbands had a higher mean age at death than their wives. The
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5 average age differences between the husbands and wives for all cohorts are
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7 shown in Table 1. The number of remarried wives was far smaller than the
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9 number of remarried husbands (n = 1,943 widowed husbands vs 613 widowed
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11 wives). The number and proportion of remarried husbands and wives were
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13 increased in the more contemporary cohorts (see Table 1). In contrast to other
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15 populations from the 19th and early 20th centuries,⁴ the majority of widowed
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17 husbands did not remarry, making it interesting to study the effect of remarriage
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19 in the Amish population.
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24 **Hazard Ratios**

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26 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
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28 associated with widowhood are displayed in Figure 1 according to birth cohort.
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30 These hazard ratios were estimated using the CPH model and design provided in
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32 Table 2a. As expected, widowhood was associated with increased mortality for
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34 both husbands and wives following the death of spouse, with HRs ranging
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36 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
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38 husbands in cohort 1850-1875). The impact of widowhood was
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40 disproportionately greater on surviving husbands than on surviving wives in the
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42 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
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44 between widowed husbands vs wives in the 1850-1875 and 1901-1925 cohorts.
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46 There was no clear trend showing that the hazard ratios changed across cohorts
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48 in either surviving husbands or wives. Since no clear differences were observed
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3 between birth cohorts, all birth cohorts were combined to estimate the association
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5 between age at widowhood and mortality.
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8 Figure 2 shows the hazard ratios (and 95% CIs) for the model in which age at
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10 widowhood is accounted for (design model in Table 2b). These results reveal
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12 markedly higher bereavement effects in each age at widowhood group compared
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14 to the results shown in Figure 1, consistent with the notion that the bereavement
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16 effect may be diminished over time and most pronounced in the years proximal to
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18 the death of the spouse.^{2 5 20 21} In nearly all age at widowhood categories, the
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20 bereavement effect is stronger in widowed husbands than in widowed wives.
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24 Number of surviving children (> 6 vs ≤ 2) was included as a covariate in the
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26 models whose results are shown in Figure 2. In general, there was a very weak
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28 association between number of surviving children and mortality in widowed
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30 husbands and widowed wives. Contrary to our expectations and a prior study,⁸ in
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32 each case, a higher number of surviving children was associated with higher
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34 mortality in the widowed husband/wife. Further, the results in Figure 2 show that
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36 the effect of bereavement decreases if the widowed individual remarries.
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40 Hazard ratios and 95% CIs for the time since bereavement analysis are shown
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42 in Figure 3. These results were obtained using the CPH model and the design
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44 defined in Table 2c. The results show that there is an increase in risk of mortality
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46 for recently widowed husbands and wives, and the hazard decreases with time
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48 since bereavement but remains significantly greater than 1. Further, the hazard is
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50 higher (not significant) in wives vs husbands during the first 12 months following
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3 bereavement.
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5 Graphical checks of the overall adequacy of the CPH models were
6 performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a
7 reasonable fit to the data. Further, the *deviance residual* plots showed that there
8 were no obvious outliers in the data.
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14 15 16 17 **DISCUSSION** 18

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21 This is the first comprehensive study to evaluate the relationship between
22 bereavement and social support in the Amish population. This study provides
23 evidence that Amish widows and widowers have increased mortality risk
24 compared to married cohort members. Although it is difficult to determine
25 whether this effect is of equivalent magnitude as that observed in studies of other
26 populations, the most recent studies of the bereavement effect using the CPH
27 suggest that our findings are generally consistent with data from other
28 populations.⁴
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40 Several previous studies on American, European, and Middle Eastern
41 populations have found that mortality is magnified in individuals widowed at a
42 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
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48 The LDS study is closest to our study because of the large family sizes and the
49 population selected as a religious isolate.⁸ The effects of bereavement on
50 mortality with respect to gender and the age at widowhood ranges observed in the
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3 Amish are also largely consistent with those observed in the LDS population,⁸ and
4 other populations such as Finland and Israel (Figure 2).^{6,22} As expected, widowers
5 showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure
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13 In the present study, the association between bereavement and mortality is
14 greater in the first 6 months for both men and women (Figure 3), consistent with
15 previous findings.^{2,3,5,20,21,23} The mortality risks in the first 6 months are lower in
16 the Amish (Figure 3) compared to some studies,^{2,5,20,21} but not all studies.^{3,23} One
17 common pattern observed in this and other studies is that the initially high
18 bereavement effect first decreases but then increases with time since
19 bereavement.^{3,23} We speculate that the increased mortality during the first 6
20 months might reflect acute effects related to the loss of a spouse, while the
21 gradual increases in mortality emerging in later life might reflect decreased
22 survival from aging-related diseases that is unmasked in the absence of spousal
23 support.
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39 Two strengths of our study are that, unlike many of the previously published
40 studies,^{5,13,16,20,21,24} we did not need to incorporate censoring methods into our
41 analysis because of the near completeness of birth and death dates of the Amish
42 widows and widowers; and we could study the effect of remarriage because there
43 were substantial numbers of widowed individuals who did and did not get
44 remarried (Table 1). Shor et al. noted that the difficulty in studying the effect of
45 remarriage in populations where all individuals are deceased because “In previous
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3 decades, widowed men almost always remarried".⁴ As suggested by previous
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5 studies,^{8 9} remarriage after the death of a spouse significantly influences the
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7 bereavement effect because it is associated with increased survival of both
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9 widowers and widows across all cohorts and age at widowhood ranges. This
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11 association is likely influenced by the fact that widowers or widows who get
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13 remarried are in sufficiently good physical and mental health to do so. Further,
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15 the survival rate is higher for widowers as compared to widows, possibly
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17 reflecting support in Amish society for males getting remarried.
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23 Interestingly, increasing numbers of surviving children at the time of
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25 widowhood did not confer a survival advantage for Amish widows or widowers.
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27 In fact, the hazard ratio was greater than 1.0 for all widowers and widows with
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29 number of surviving children > 6 as compared to ≤ 2 . This result was counter to
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31 our hypothesis that children can help provide social support for their parents. The
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33 lack of protective association was similarly observed when the number of
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35 surviving children was considered as a linear or as a categorical variable (data not
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37 shown). This contrasts with data from the Utah Population Database, in which
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39 increasing numbers of children were associated with a decreased hazard ratio.⁸
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44 The implication of these findings is that differential familial support
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46 following bereavement does not appear to be the key factor affecting mortality
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48 increases in widowed Amish populations. One potential explanation is that
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50 spouses, as "attachment figures," provide a unique, irreplaceable social support
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52 which must be considered independently from other ancillary support providers,
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3 such as children.²⁵ Data suggesting that relative mortality risk is also elevated in
4 divorced and never-married populations may support this hypothesis.³ In addition,
5 quality of social support has significantly greater effects on well-being than
6 quantity of support in elderly populations of both sexes.²⁶
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12 In adults aged 65 and older, lower reported social support has been associated
13 with decreased life satisfaction and increased depressive symptoms.²⁷ Family
14 members and close friends should be especially vigilant during the sensitive acute
15 period following the loss of a spouse, when relative mortality risk is the highest.
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22 A limitation of our data is that causes of death were unavailable. Future
23 research is needed to determine whether differential measures of social support
24 may be associated with certain causes of death in widowed populations. Divorce
25 in the Amish is sufficiently rare that this was not a major potential source of error.
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32 Our study results indicate that remarriage plays an important role in
33 improving the survival rate of widows and widowers in the Amish population.
34 Contrary to results from previous studies, an increase in the number of surviving
35 children was associated with a decreased survival rate of widowed individuals.
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44 **Contributors** BDM conceived the study. AS and SS did the research and data
45 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
46 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
47 managed the project with assistance from BDM. AAS manages the AGDB project
48 at the National Institutes of Health and KAR managed the AGDB data at
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3 University of Maryland Medical School during the period of this project. ARS
4 provided resources and advice. All authors proofread and approved the
5 manuscript.
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9
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14 Geriatric Research and Education Center, Baltimore Veterans Administration
15 Medical Center.
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20 **Competing interests** None.
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25 **Ethics approval** The construction and maintenance of AGDB are covered under
26 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
27 Investigator.
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35 **Provenance and peer review** Not commissioned; externally peer reviewed.
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40 **Data sharing statement** Data from the AGDB are available to investigators
41 (including ARS and various others not participating in this study) who have an
42 IRB-approved protocol to study the Amish or other Anabaptist groups.
43 Investigators can request access to AGDB by writing to AAS and to Dr.
44 Biesecker.
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FIGURE LEGENDS

Figure 1. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a).

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).

TABLES

Table 1: Characteristics of 15,611 spouse pairs according to birth cohort of husband

	Cohorts			
	Pre-1850	1850-1875	1876-1900	1901-1925
Number of couples	3711	3210	4466	4224
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2360(53.0)	2439(57.7)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	2106(47.0)	1785(42.3)
Mean husband age at widowhood	63.0	59.8	62.2	67.9
Mean wife age at widowhood	62.0	62.8	66.5	69.6
Mean widowed husband survival in years	14.4	18.3	18.6	15.0
Mean widowed wife survival in years	15.3	16.0	17.0	15.1
Mean husband age at death	71.1	72.0	74.6	76.2
Mean wife age at death	69.6	68.5	72.7	77.0
Mean age difference husband-wife	3.4	2.8	2.1	1.3
Mean number of children	5.4	5.3	5.3	5.8
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	13(33.8)	588(33.0)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	221(9.4)	194(8.0)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0

ID ₁	51	52	0	0	0	0	0	0	1	0
ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

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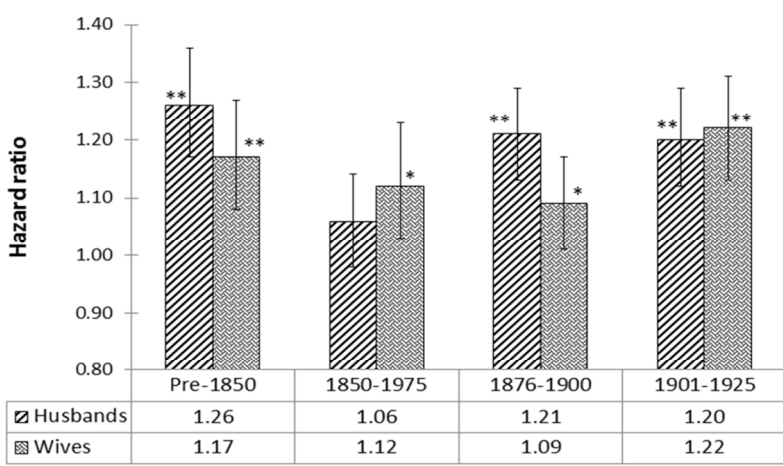


Figure 1. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a).
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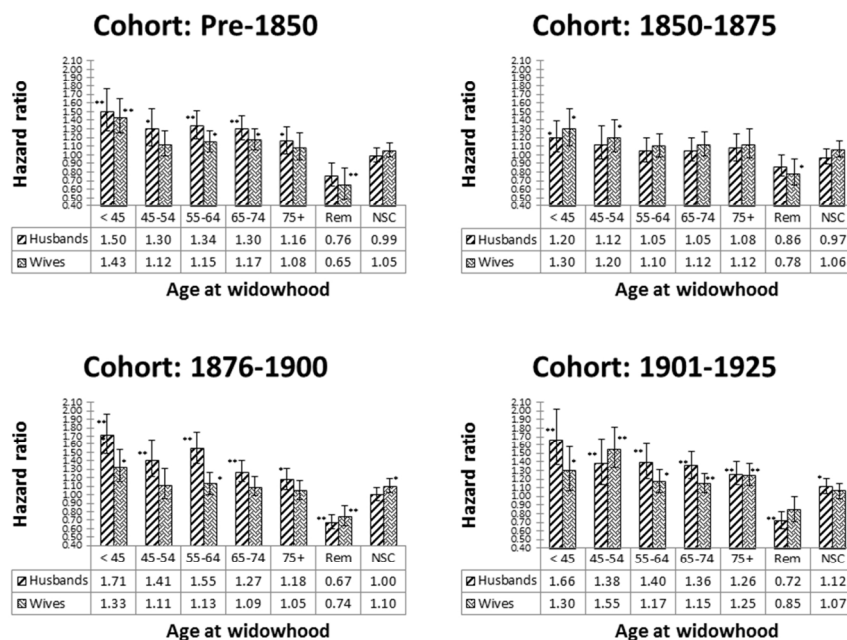


Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).
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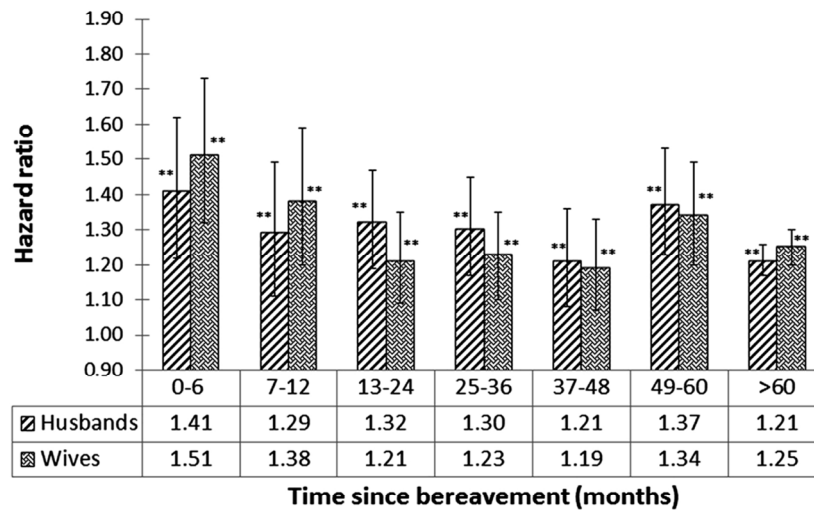


Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).
173x233mm (300 x 300 DPI)

STROBE Statement—Checklist of items that should be included in reports of *cohort studies*

	Item No	Recommendation
Title and abstract	1	<p>(a) Indicate the study's design with a commonly used term in the title or the abstract (From Abstract) This study investigates the association between bereavement and the mortality of a surviving spouse among 15,611 Amish couples born during 1725-1925. We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using the Cox proportional hazard model.</p> <p>(b) Provide in the abstract an informative and balanced summary of what was done and what was found. See Abstract for findings.</p>
Introduction		
Background/rationale	2	<p>Explain the scientific background and rationale for the investigation being reported (From Introduction) It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the "bereavement effect," is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.²⁻⁵ Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.⁶⁻⁷ To gain a different perspective on potential influences of social support on bereavement, we assessed the bereavement effect in an Amish population, whose culture is characterized by its core beliefs in community and social cohesion. Examples of the strong degree of social support within the Amish include community-managed health insurance and community ties through membership in the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹² We reasoned that the high level of social support in the Amish population might mitigate the bereavement effect.</p>
Objectives	3	<p>State specific objectives, including any prespecified hypotheses (From Abstract) We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support. We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using the Cox proportional hazard model.</p>
Methods		
Study design	4	<p>Present key elements of study design early in the paper (From Introduction) We used Cox proportional hazard (CPH) models to analyze the association of bereavement and mortality of widowed husbands and wives. We considered remarriage and number of surviving children as additional potential modifiers of the bereavement effect. (From Material and Methods). The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement of the Amish population.¹⁴⁻¹⁵</p>
Setting	5	<p>Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection (From Material and Methods) The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement on the Amish population.¹⁴⁻¹⁵ The AGDB is a computerized genealogy database of Amish and some Mennonite populations, last updated in 2010. The AGDB contains information for 539,822 Amish individuals and 136,213 Amish couples. This database includes information about the family relationships and the birth, marriage, and death dates of children of Amish individuals</p>

		from North America, mostly located in Pennsylvania, Ohio, and Indiana.
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up (From Materials and Methods) For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1926; (2) their birth and death dates were recorded; (3) they were in their first marriage; and (4) they did not die on the same day. Constraint (4) excludes couples who may have died from a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 31, 1925. Our data set included 15,611 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood.
		(b) For matched studies, give matching criteria and number of exposed and unexposed This is not a matched study
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable See Material and Methods, especially Table 2.
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group (From Material and Methods) The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement on the Amish population. ^{14 15} The AGDB is a computerized genealogy database of Amish and some Mennonite populations, last updated in 2010. The AGDB contains information for 539,822 Amish individuals and 136,213 Amish couples. This database includes information about the family relationships and the birth, marriage, and death dates of children of Amish individuals from North America, mostly located in Pennsylvania, Ohio, and Indiana.
Bias	9	Describe any efforts to address potential sources of bias We excluded couples who died on the same day. The data are not censored.
Study size	10	Explain how the study size was arrived at All couples in AGDB meeting the criteria described in item 5.
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why (From Material and Methods) For some analyses, these couples were first partitioned into eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-1900, and 1901-1925) and sex. For the time since bereavement analyses, we did not partition into cohorts because the number of widow(er)s who died within 6 months after being bereaved is small (N=224). Characteristics of these couples by husband birth cohort are shown in Table 1.
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding See Material and Methods, especially Table 2.
		(b) Describe any methods used to examine subgroups and interactions (From Material and Methods) For some analyses, these couples were first partitioned into eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-1900, and 1901-1925) and sex.

		(c)	Explain how missing data were addressed Couples with missing dates were excluded.
		(d)	If applicable, explain how loss to follow-up was addressed Not applicable.
		(e)	Describe any sensitivity analyses Not applicable
Results			
Participants	13*	(a)	Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed See Table 1
		(b)	Give reasons for non-participation at each stage Not applicable
		(c)	Consider use of a flow diagram We preferred to describe the analysis designs in Table 2.
Descriptive data	14*	(a)	Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders See Table 1.
		(b)	Indicate number of participants with missing data for each variable of interest Couples with missing dates were excluded.
		(c)	Summarise follow-up time (eg, average and total amount) This is a retrospective study; All participants are deceased.
Outcome data	15*		Report numbers of outcome events or summary measures over time See Table 1.
Main results	16	(a)	Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included See Figures 1-3 and text of Results. Figures do show 95% confidence intervals.
		(b)	Report category boundaries when continuous variables were categorized See Table 2.
		(c)	If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period Not applicable.
Other analyses	17		Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses See Results text.

Discussion		
Key results	18	<p>Summarise key results with reference to study objectives</p> <p>(From Discussion) This study provides evidence that Amish widows and widowers have increased mortality risk compared to married cohort members. Although it is difficult to determine whether this effect is of equivalent magnitude as that observed in studies of other populations, the most recent studies of the bereavement effect using the CPH suggest that our findings are generally consistent with data from other populations.⁴</p> <p>Interestingly, increasing numbers of surviving children at the time of widowhood did not confer a survival advantage for Amish widows or widowers. In fact, the hazard ratio was greater than 1.0 for all widowers and widows with number of surviving children > 6 as compared to ≤ 2. This result was counter to our hypothesis that children can help provide social support for their parents.</p>
Limitations	19	<p>Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias</p> <p>(From Discussion) A limitation of our data is that causes of death were unavailable. Future research is needed to determine whether differential measures of social support may be associated with certain causes of death in widowed populations. Divorce in the Amish is sufficiently rare that this was not a major potential source of error.</p>
Interpretation	20	<p>Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence</p> <p>(From Discussion) The effects of bereavement on mortality with respect to gender and the age at widowhood ranges observed in the Amish are also largely consistent with those observed in the LDS population,⁸ and other populations such as Finland and Israel (Figure 2).^{6,22} As expected, widowers showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure 1).</p> <p>Two strengths of our study are that, unlike many of the previously published studies,^{5,13,16,20,21,24} we did not need to incorporate censoring methods into our analysis because of the near completeness of birth and death dates of the Amish widows and widowers; and we could study the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1).</p> <p>Our study results indicate that remarriage plays an important role in improving the survival rate of widows and widowers in the Amish population. Contrary to results from previous studies, an increase in the number of surviving children was associated with a decreased survival rate of widowed individuals.</p>
Generalisability	21	<p>Discuss the generalisability (external validity) of the study results</p> <p>(From Discussion) One common pattern observed in this and other studies is that the initially high bereavement effect first decreases but then increases with time since bereavement.^{3,23} We speculate that the increased mortality during the first 6 months might reflect acute effects related to the loss of a spouse, while the gradual increases in mortality emerging in later life might reflect decreased survival from aging-related diseases that is unmasked in the absence of spousal support.</p> <p>As suggested by previous studies,^{8,9} remarriage after the death of a spouse significantly influences the bereavement effect because it is associated with increased survival of both widowers and widows across all cohorts and age at widowhood ranges. This association is likely influenced by the fact that widowers or widows who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for widowers as compared to widows, possibly reflecting support in Amish society for males getting remarried.</p>

1 Our study results indicate that remarriage plays an important role in improving the
2 survival rate of widows and widowers in the Amish population.
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Other information

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5 Funding 22 Give the source of funding and the role of the funders for the present study and, if
6 applicable, for the original study on which the present article is based
7 (From Acknowledgments) This work was supported by research grants R01 HL69313,
8 R01 DK54261, R01 AG1872801, R01 HL088119, R01 AR046838, and U01 HL72515
9 from the National Institutes of Health. This research was supported in part by the
10 Intramural Research Program of the National Institutes of Health, NLM and the
11 Geriatric Research and Education Center, Baltimore Veterans Administration Medical
12 Center. These funders had no role in the study itself.
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16 *Give information separately for exposed and unexposed groups.
17

18 **Note:** An Explanation and Elaboration article discusses each checklist item and gives methodological background and
19 published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely
20 available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at
21 <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is
22 available at <http://www.strobe-statement.org>.
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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Secondary Subject Heading:	Geriatric medicine, Public health, Sociology
Keywords:	EPIDEMIOLOGY, GERIATRIC MEDICINE, MENTAL HEALTH, PUBLIC HEALTH

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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Author-suggested Keywords: bereavement effect, Amish, remarriage, social support, Cox proportional hazards, time-dependent covariate

Word Counts:

Abstract: 248

Introduction through Discussion: 3,377.

ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 15,611 Amish couples born during 1725-1925 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure The survival time is 'age'; event is 'death'. Hazard ratios of widowed individuals with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both widows and widowers. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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3 socially cohesive Amish community. Remarriage is associated with a significant
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5 decrease in the mortality risk among widowed individuals. Contrary to results
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8 from previous studies, an increase in the number of surviving children was
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10 associated with decreased survival rate.
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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.
- We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks of the bereaved individuals and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of a large and uncensored dataset, our hazard ratio estimates are mostly significant with narrow confidence intervals.
- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.

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- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.

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INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.^{2 5} Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6 7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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The Amish maintain a cultural identity distinct from mainstream American

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3 culture that is characterized by their traditional dress, a plain lifestyle, and non-
4 adoption of modern technology (e.g., electricity, cars, telephones), German
5 dialect, separate school system, and ultra-conservative Anabaptist religious
6 practices. A central tenet of Amish culture throughout their history in the USA
7 has been social cohesiveness with emphasis on family and community. Members
8 of this tight-knit society have extraordinary social support from cradle to grave,
9 including community-managed health insurance and support during times of need.
10 Elderly parents are taken care of by their children and neighbors; they do not use
11 assisted living or nursing homes to care for their elderly.
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24 To gain a different perspective on potential influences of social support on
25 bereavement, we assessed the bereavement effect in an Amish population, whose
26 culture is characterized by its core beliefs in community and social cohesion.
27 Examples of the strong degree of social support within the Amish include
28 community-managed health insurance and community ties through membership in
29 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
30 We reasoned that the high level of social support in the Amish population might
31 mitigate the bereavement effect. Previous studies used covariates such as
32 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
33 neighborhood interaction, and household size to study the relationship between
34 bereavement and social support.^{4 8 13}
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50 We estimated bereavement effects in widows and widowers separately, and
51 further assessed mortality risks as a function of age at widowhood and time since
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3 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
4 association of bereavement and mortality of widowed husbands and wives. We
5 considered remarriage and number of surviving children as additional potential
6 modifiers of the bereavement effect.
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12 13 14 15 **MATERIAL AND METHODS**

16 17 **Data source**

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19 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
20 effect of bereavement on the Amish population.^{14 15} The AGDB is a
21 computerized genealogy database of Amish and some Mennonite populations, last
22 updated in 2010. This database includes information about the family
23 relationships and the birth, marriage, and death dates of children of Amish
24 individuals from North America, mostly located in Pennsylvania, Ohio, and
25 Indiana. The “individual table” of AGDB contains information about 539,822
26 individuals. The “relationship table” includes information about 136,213 Amish
27 couples. An individual who is married multiple times participates in multiple
28 relationship table entries. There are 1,369 relationship entries among the 136,213
29 entries concerning children for whom one or both biological parents are unknown
30 (Figure 1).
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48 Amish people rarely give birth out of wedlock, and a vast majority of the
49 mating samples we used are documented as marriages in the three original sources
50 from which AGDB is derived. However, there is less availability of marriage
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3 dates than birth dates; we did not wish to exclude couples with missing marriage
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5 dates. For this study, we included couples in which both spouses met four
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7 conditions: (1) they were born prior to January 1, 1926; (2) their birth and
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9 death dates were recorded; (3) they were in their first marriage; and (4) they did
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11 not die on the same day. Constraint (4) excludes couples who may have died from
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13 a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion
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15 criteria. The oldest person included was born on February 14, 1725, and the
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17 youngest on December 31, 1925. See Figure 1 for how the exclusion criteria were
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19 applied.
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25 Our data set included 15,611 couples with information on their date of birth,
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27 date of death, number of surviving children, family IDs, and remarriage status
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29 after widowhood. For some analyses, these couples were first partitioned into
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31 eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-
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33 1900, and 1901-1925) and sex. For the time since bereavement analyses, we did
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35 not partition into cohorts because the number of widow(er)s who died within 6
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37 months after being bereaved is small (N=224). Characteristics of these couples by
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39 husband birth cohort are shown in Table 1.
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44 45 **Statistical analyses**

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47 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
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49 where the response variable or survival time 'age at widowhood or death' and
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51 the event is 'death'. The model is used to study the association of widowhood
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53 and mortality rates in the surviving spouse, while adjusting for covariates such as
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3 education, health habits, age in years, number of children, and remarriage. In some
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5 of our analyses, we adjusted for remarriage and number of children as
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7 covariates; we did not adjust for education or health habits. Widows and
8
9 widowers were always analyzed separately.
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13 The main covariate, widowhood, was monitored as a time-dependent
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15 covariate, by assigning a value of 1 for each time period the individual was
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17 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
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19 covariate, by assigning a value 1 for the widowed period if the individual
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21 remarried and 0 otherwise. The number of surviving children was a time-
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23 independent covariate and divided into categories, as follows: 0=number of
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25 surviving children is ≤ 2 , 1=number of surviving children is between 3 and 6, and
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27 2=number of surviving children is > 6 . The categories ≤ 2 children, 3-6 children, and
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29 > 6 children are separate. These boundaries were chosen to ensure categories that were
30
31 roughly balanced in size.
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36 Survival analyses were performed using the R programming environment.¹⁷
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38 All estimates and confidence intervals were obtained using the `coxph` function
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40 available in the `survival` package.
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43 **Representation of survival data for CPH analysis**

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45 We show below examples of how we represented the survival data for CPH
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47 analysis using a time modeling approach. To represent the survival data columns, the
48
49 standard approach is to convert the couple's demographic information, date of
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51 death, date of birth, remarriage and number of surviving children into columns
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3 representing **start** time, **stop** time, **event** status, and all of the included covariates
4 (widowhood, remarriage and number of surviving children).^{18 19} Here (**start**,
5
6 **stop**] is an interval of risk, open on the left and closed on the right; the **event**
7
8 column is set to 1 if the subject had an event (death) at time **stop** and is 0
9
10 otherwise. To illustrate the subtleties, we consider a hypothetical example of
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12 three wives (ID1, ID2 and ID3) from the cohort 1850-1875.
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16 • *ID*1: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
17
18 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
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- 21
22 • *ID*2: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
23
24 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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- 27
28 • *ID*3: born on 03/01/1871; never widowed and never remarried; number of
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30 surviving children = 4 and died on 04/01/1930 at age 59.
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42 The data used for the CPH considering widowhood as a single time-
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44 dependent covariate is presented in Table 2a. The following model was used to
45
46 estimate the association between widowhood and mortality.
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$$48 \quad h(t) = h_0(t) \exp[\beta W]$$

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50 This model can be run with additional covariates and results that adjust for the
51
52 ability to remarry and the number of surviving children are presented.
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Timing of widowhood

The above model considers widowhood as a single time-dependent covariate, evaluating the association between mortality and being widowed. One can further ask if that association varies over the life course by considering the impact of age at widowhood on mortality. We addressed this issue following the approach of Mineau et al. by adding terms for age at widowhood into the model as covariates (<45, 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to age at widowhood (see Table 2b).⁸ These terms were included in the model as time-dependent dummy variables, created to represent the widowhood experience of each individual across the 5 age windows spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} . Further, we also addressed the impact of remarriage and number of surviving children on mortality of widowed individuals.

The following model can be used to estimate the association between each of the five dummy variables and mortality:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , W_{75+} columns provided in the Table 2b.

Time since bereavement

Next, we evaluated the association between mortality and time since bereavement or widowhood. We followed the approach of Schaefer et al. by considering the

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3 following time since bereavement ranges: 0-6 months, 7-12 months, 13-24
4 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This
5 approach allowed us to estimate HR according to time since bereavement (see
6 Table 2c). The columns TSB₀₋₆, TSB₇₋₁₂, ... in Table 2c are time dependent
7 covariates that change with the survival time associated with widowed husbands
8 and wives. We did not account for remarriage in this analysis because of missing
9 remarriage dates.

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20 The following model was used to study the association between mortality and
21 time since bereavement:

$$22 \quad h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

23
24 where W_1, \dots, W_7 dummy variables are associated with the TSB₀₋₆, TSB₇₋₁₂,
25 TSB₁₃₋₂₄, TSB₂₅₋₃₆, TSB₃₇₋₄₈, TSB₄₉₋₆₀ and TSB_{>60} columns provided in
26 the Table 2c.
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37 RESULTS

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40 We initially partitioned the 15,611 couples from AGDB into four cohorts
41 ranging in size from 3,210 (husband birth year 1850-1875) to 4,466 (husband
42 birth year 1876-1900). Wives were more likely to die after their husbands in all
43 cohorts, although in the three earliest cohorts (prior to 1850, 1850-1875, and
44 1876-1900), husbands had a higher mean age at death than their wives. The
45 average age differences between the husbands and wives for all cohorts are
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3 shown in Table 1. The number of remarried wives was far smaller than the
4 number of remarried husbands (n = 1,943 widowed husbands vs 613 widowed
5 wives). The number and proportion of remarried husbands and wives were
6 increased in the more contemporary cohorts (see Table 1). In contrast to other
7 populations from the 19th and early 20th centuries,⁴ the majority of widowed
8 husbands did not remarry, making it interesting to study the effect of remarriage
9 in the Amish population.
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19 **Hazard Ratios**

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21 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
22 associated with widowhood are displayed in Figure 2 according to birth cohort.
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24 These hazard ratios were estimated using the CPH model and design provided in
25 Table 2a. As expected, widowhood was associated with increased mortality for
26 both husbands and wives following the death of spouse, with HRs ranging
27 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
28 husbands in cohort 1850-1875). The impact of widowhood was
29 disproportionately greater on surviving husbands than on surviving wives in the
30 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
31 between widowed husbands vs wives in the 1850-1875 and 1901-1925 cohorts.
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33 There was no clear trend showing that the hazard ratios changed across cohorts
34 in either surviving husbands or wives. Since no clear differences were observed
35 between birth cohorts, all birth cohorts were combined to estimate the association
36 between age at widowhood and mortality.
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Figure 3 shows the hazard ratios (and 95% CIs) for the model in which age at widowhood is accounted for (design model in Table 2b). These results reveal markedly higher bereavement effects in each age at widowhood group compared to the results shown in Figure 2, consistent with the notion that the bereavement effect may be diminished over time and most pronounced in the years proximal to the death of the spouse.^{2 5 20 21} In nearly all age at widowhood categories, the bereavement effect is stronger in widowed husbands than in widowed wives.

Number of surviving children (> 6 vs ≤ 2) was included as a covariate in the models whose results are shown in Figure 3. In general, there was a very weak association between number of surviving children and mortality in widowed husbands and widowed wives. Contrary to our expectations and a prior study,⁸ in each case, a higher number of surviving children was associated with higher mortality in the widowed husband/wife. When analyses were repeated without the number of surviving children as a covariate, the estimates of the hazard ratios were essentially unchanged (data not shown). Further, the results in Figure 3 show that the effect of bereavement decreases if the widowed individual remarries.

Hazard ratios and 95% CIs for the time since bereavement analysis are shown in Figure 4. These results were obtained using the CPH model and the design defined in Table 2c. The results show that there is an increase in risk of mortality for recently widowed husbands and wives, and the hazard decreases with time since bereavement but remains significantly greater than 1. Further, the hazard is higher (not significant) in wives vs husbands during the first 12 months following

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3 bereavement. To address the issue that many individuals in the 1901-1925 cohort
4 may still have been alive at the last AGDB update and hence censored, we
5 performed the analysis for time since bereavement omitting the cohort 1901-1925.
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8 The results show that the hazard ratios provided in Figure 4 have not changed
9 significantly; for example, for 25-26 months post bereavement the hazard ratios
10 change from 1.30 and 1.23 for all cohort to 1.25 and 1.17 for the three eldest
11 cohorts.
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20 Graphical checks of the overall adequacy of the CPH models were
21 performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a
22 reasonable fit to the data (data not shown). The *deviance residual* plots
23 revealed no obvious outliers in the data (data not shown). Further, the Wald test
24 statistic was used to test the fit of the final model,¹⁸ and according to this test
25 statistic, the final model fits the data reasonably well (Supplementary Tables S1,
26 S2, and S3).
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39 DISCUSSION

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42 This is the first comprehensive study to evaluate the relationship between
43 bereavement and social support in the Amish population. This study provides
44 evidence that Amish widows and widowers have increased mortality risk
45 compared to married cohort members. Although it is difficult to determine
46 whether this effect is of equivalent magnitude as that observed in studies of other
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3 populations, the most recent studies of the bereavement effect using the CPH
4 suggest that our findings are generally consistent with data from other
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6 populations.⁴
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10 Several previous studies on American, European, and Middle Eastern
11 populations have found that mortality is magnified in individuals widowed at a
12 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
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14 The LDS study is closest to our study because of the large family sizes and the
15 population selected as a religious isolate.⁸ The effects of bereavement on
16 mortality with respect to gender and the age at widowhood ranges observed in the
17 Amish are also largely consistent with those observed in the LDS population,⁸ and
18 other populations such as Finland and Israel (Figure 3).^{6 22} As expected, widowers
19 showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure
20 2).
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34 In the present study, the association between bereavement and mortality is
35 greater in the first 6 months for both men and women (Figure 4), consistent with
36 previous findings.^{2 3 5 20 21 23} The mortality risks in the first 6 months are lower in
37 the Amish (Figure 4) compared to some studies,^{2 5 20 21} but not all studies.^{3 23} One
38 common pattern observed in this and other studies is that the initially high
39 bereavement effect first decreases but then increases with time since
40 bereavement.^{3 23} We speculate that the increased mortality during the first 6
41 months might reflect acute effects related to the loss of a spouse, while the
42 gradual increases in mortality emerging in later life might reflect decreased
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3 survival from aging-related diseases that is unmasked in the absence of spousal
4 support.
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8 Two strengths of our study are that, unlike many of the previously published
9 studies,^{5 13 16 20 21 24} we did not incorporate censoring methods into our analysis
10 because of the high availability of death dates of the Amish widows and widowers
11 in the first three cohorts; and we could study the effect of remarriage because
12 there were substantial numbers of widowed individuals who did and did not get
13 remarried (Table 1). Shor et al. noted that the difficulty in studying the effect of
14 remarriage in populations where all individuals are deceased because “In previous
15 decades, widowed men almost always remarried”.⁴ As suggested by previous
16 studies,^{8 9} remarriage after the death of a spouse significantly influences the
17 bereavement effect because it is associated with increased survival of both
18 widowers and widows across all cohorts and age at widowhood ranges. This
19 association is likely influenced by the fact that widowers or widows who get
20 remarried are in sufficiently good physical and mental health to do so. Further,
21 the survival rate is higher for widowers as compared to widows, possibly
22 reflecting support in Amish society for males getting remarried.
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43 Interestingly, increasing numbers of surviving children at the time of
44 widowhood did not confer a survival advantage for Amish widows or widowers.
45 This result was counter to our hypothesis that children can help provide social
46 support for their parents. The hazard ratio was greater than 1.0 (but not
47 significant) for all widowers and widows with number of surviving children > 6
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3 as compared to ≤ 2 . Spouses in the Amish society may also provide unique
4 emotional, psychological, and social support to each other which cannot be
5 provided by their surviving children. The lack of protective association was
6 similarly observed when the number of surviving children was considered as a
7 linear or as a categorical variable (data not shown). This contrasts with data from
8 the Utah Population Database, in which increasing numbers of children were
9 associated with a decreased hazard ratio.⁸
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22 The implication of these findings is that differential familial support
23 following bereavement does not appear to be the key factor affecting mortality
24 increases in widowed Amish populations. One potential explanation is that
25 spouses, as “attachment figures,” provide a unique, irreplaceable social support
26 which must be considered independently from other ancillary support providers,
27 such as children.²⁵ Data suggesting that relative mortality risk is also elevated in
28 divorced and never-married populations may support this hypothesis.³ In addition,
29 quality of social support has significantly greater effects on well-being than
30 quantity of support in elderly populations of both sexes.²⁶
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44 In adults aged 65 and older, lower reported social support has been associated
45 with decreased life satisfaction and increased depressive symptoms.²⁷ Family
46 members and close friends should be especially vigilant during the sensitive acute
47 period following the loss of a spouse, when relative mortality risk is the highest.
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52 A limitation of our data is that causes of death were unavailable. Future
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3 research is needed to determine whether differential measures of social support
4 may be associated with certain causes of death in widowed populations. Divorce
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6 in the Amish is sufficiently rare that this was not a major potential source of error.
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10 Our study results indicate that remarriage plays an important role in
11 improving the survival rate of widows and widowers in the Amish population.
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13 Contrary to results from previous studies, an increase in the number of surviving
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15 children was associated with a decreased survival rate of widowed individuals.
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20 to improve the manuscript.
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24
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26 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
27 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
28 managed the project with assistance from BDM. AAS manages the AGDB project
29 at the National Institutes of Health and KAR managed the AGDB data at
30 University of Maryland Medical School during the period of this project. ARS
31 provided resources and advice. All authors proofread and approved the
32 manuscript.
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3 Medical Center.
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6 **Competing interests** None.
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10 **Ethics approval** The construction and maintenance of AGDB are covered under
11 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
12 Investigator.
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17 **Provenance and peer review** Not commissioned; externally peer reviewed.
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21 **Data sharing statement** Data from the AGDB are available to investigators
22 (including ARS and various others not participating in this study) who have an
23 IRB-approved protocol to study the Amish or other Anabaptist groups.
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25 Investigators can request access to AGDB by writing to AAS and to Dr.
26 Biesecker.
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FIGURE LEGENDS

Figure 1. A flow diagram which represents all the steps performed for filtering 15,611 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. “Unknown spouse” refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. “Birth year too late” means that the birth year of the husband is known and is > 1925 . “Dates not recognized by R” are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. “Implausible birth or death dates” refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a).

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).

Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).

TABLES

Table 1: Characteristics of 15,611 spouse pairs according to birth cohort of husband

	Cohorts			
	Pre-1850	1850-1875	1876-1900	1901-1925
Number of couples	3711	3210	4466	4224
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2360(53.0)	2439(57.7)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	2106(47.0)	1785(42.3)
Mean husband age at widowhood	63.0	59.8	62.2	67.9
Mean wife age at widowhood	62.0	62.8	66.5	69.6
Mean widowed husband survival in years	14.4	18.3	18.6	15.0
Mean widowed wife survival in years	15.3	16.0	17.0	15.1
Mean husband age at death	71.1	72.0	74.6	76.2
Mean wife age at death	69.6	68.5	72.7	77.0
Mean age difference husband-wife	3.4	2.8	2.1	1.3
Mean number of children	5.4	5.3	5.3	5.8
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	13(33.8)	588(33.0)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	221(9.4)	194(8.0)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0

ID ₁	51	52	0	0	0	0	0	0	1	0
ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 15,611 Amish couples born during 1725-1925 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure [The survival time is 'age'; event is 'death'](#). Hazard ratios of [widowed individuals](#) with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both widows and widowers. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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socially cohesive Amish community. Remarriage is associated with a significant decrease in the mortality risk among widowed individuals. Contrary to results from previous studies, an increase in the number of surviving children was associated with decreased survival rate.

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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.
- We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks of the bereaved individuals and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of a large and uncensored dataset, our hazard ratio estimates are mostly significant with narrow confidence intervals.
- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.

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- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.

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INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.²⁻⁵ Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6,7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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[The Amish maintain a cultural identity distinct from mainstream American](#)

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9 culture that is characterized by their traditional dress, a plain lifestyle, and non-
10 adoption of modern technology (e.g., electricity, cars, telephones), German
11 dialect, separate school system, and ultra-conservative Anabaptist religious
12 practices. A central tenet of Amish culture throughout their history in the USA
13 has been social cohesiveness with emphasis on family and community. Members
14 of this tight-knit society have extraordinary social support from cradle to grave,
15 including community-managed health insurance and support during times of need.
16 Elderly parents are taken care of by their children and neighbors; they do not use
17 assisted living or nursing homes to care for their elderly.

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26 To gain a different perspective on potential influences of social support on
27 bereavement, we assessed the bereavement effect in an Amish population, whose
28 culture is characterized by its core beliefs in community and social cohesion.
29 Examples of the strong degree of social support within the Amish include
30 community-managed health insurance and community ties through membership in
31 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
32 We reasoned that the high level of social support in the Amish population might
33 mitigate the bereavement effect. Previous studies used covariates such as
34 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
35 neighborhood interaction, and household size to study the relationship between
36 bereavement and social support.^{4 8 13}

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46 We estimated bereavement effects in widows and widowers separately, and
47 further assessed mortality risks as a function of age at widowhood and time since
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9 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
10 association of bereavement and mortality of widowed husbands and wives. We
11 considered remarriage and number of surviving children as additional potential
12 modifiers of the bereavement effect.
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16 17 18 **MATERIAL AND METHODS**

19 **Data source**

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21 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
22 effect of bereavement on the Amish population.^{14 15} The AGDB is a
23 computerized genealogy database of Amish and some Mennonite populations, last
24 updated in 2010. ~~The AGDB contains information for 539,822 Amish individuals
25 and 136,213 Amish couples.~~ This database includes information about the family
26 relationships and the birth, marriage, and death dates of children of Amish
27 individuals from North America, mostly located in Pennsylvania, Ohio, and
28 Indiana. The "individual table" of AGDB contains information about 539,822
29 individuals. The "relationship table" includes information about 136,213 Amish
30 couples. An individual who is married multiple times participates in multiple
31 relationship table entries. There are 1,369 relationship entries among the 136,213
32 entries concerning children for whom one or both biological parents are unknown
33 (Figure 1).
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46 Amish people rarely give birth out of wedlock, and a vast majority of the
47 mating samples we used are documented as marriages in the three original sources
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9 from which AGDB is derived. However, there is less availability of marriage
10 dates than birth dates; we did not wish to exclude couples with missing marriage
11 dates. For this study, we included couples in which both spouses met four
12 conditions: (1) they were born prior to January 1, 1926; (2) their birth and
13 death dates were recorded; (3) they were in their first marriage; and (4) they did
14 not die on the same day. Constraint (4) excludes couples who may have died from
15 a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion
16 criteria. The oldest person included was born on February 14, 1725, and the
17 youngest on December 31, 1925. [See Figure 1 for how the exclusion criteria were](#)
18 [applied.](#)
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28 Our data set included 15,611 couples with information on their date of birth,
29 date of death, number of surviving children, family IDs, and remarriage status
30 after widowhood. For some analyses, these couples were first partitioned into
31 eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-
32 1900, and 1901-1925) and sex. For the time since bereavement analyses, we did
33 not partition into cohorts because the number of widow(er)s who died within 6
34 months after being bereaved is small (N=224). Characteristics of these couples by
35 husband birth cohort are shown in Table 1.
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44 **Statistical analyses**

45 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
46 [where the response variable or survival time 'age at widowhood or death' and](#)
47 [the event is 'death'. The model is used—Hazards \(CPH\) model](#) to study the
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9 association of widowhood and mortality rates in the surviving spouse, while
10 adjusting for covariates such as education, health habits, age in years, number of
11 children, and remarriage. In some of our analyses, we adjusted for remarriage
12 and number of children as covariates; [we did not adjust for education or health](#)
13 [habits. Widows and widowers were always analyzed separately.](#)
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18 The main covariate, widowhood, was monitored as a time-dependent
19 covariate, by assigning a value of 1 for each time period the individual was
20 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
21 covariate, by assigning a value 1 for the widowed period if the individual
22 remarried and 0 otherwise. The number of surviving children was a time-
23 independent covariate and divided into categories, as follows: 0=number of
24 surviving children is ≤ 2 , 1=number of surviving children is between 3 and 6, and
25 2=number of surviving children is > 6 . [The categories \$\leq 2\$ children, 3-6 children, and](#)
26 [\$> 6\$ children are separate. These boundaries were chosen to ensure categories that were](#)
27 [roughly balanced in size.](#)
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32 Survival analyses were performed using the R programming environment.¹⁷
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34 All estimates and confidence intervals were obtained using the `coxph` function
35 available in the `survival` package.
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37 38 39 **Representation of survival data for CPH analysis**

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42 We show below examples of how we represented the survival data for CPH
43 analysis using a time modeling approach. To represent the survival data columns, the
44 standard approach is to convert the couple's demographic information, date of
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9 death, date of birth, remarriage and number of surviving children into columns
10 representing **start** time, **stop** time, **event** status, and all of the included covariates
11 (widowhood, remarriage and number of surviving children).^{18 19} Here (**start**,
12 **stop**) is an interval of risk, open on the left and closed on the right; the **event**
13 column is set to 1 if the subject had an event (death) at time **stop** and is 0
14 otherwise. To illustrate the subtleties, we consider a hypothetical example of
15 three wives (ID₁, ID₂ and ID₃) from the cohort 1850-1875.
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24 • ID₁: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
25 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
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30 • ID₂: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
31 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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35 • ID₃: born on 03/01/1871; never widowed and never remarried; number of
36 surviving children = 4 and died on 04/01/1930 at age 59.
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41 The data used for the CPH considering widowhood as a single time-
42 dependent covariate is presented in Table 2a. The following model was used to
43 estimate the association between widowhood and mortality.
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$$46 \quad h(t) = h_0(t) \exp[\beta W]$$

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49 This model can be run with additional covariates and results that adjust for the
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9 ability to remarry and the number of surviving children are presented.

10 11 **Timing of widowhood**

12 The above model considers widowhood as a single time-dependent
13 covariate, evaluating the association between mortality and being
14 widowed. One can further ask if that association varies over the life
15 course by considering the impact of age at widowhood on mortality.
16 We addressed this issue following the approach of Mineau et al. by
17 adding terms for age at widowhood into the model as covariates (<45,
18 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to
19 age at widowhood (see Table 2b).⁸ These terms were included in the model
20 as time-dependent dummy variables, created to represent the
21 widowhood experience of each individual across the 5 age windows
22 spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} .
23 Further, we also addressed the impact of remarriage and number of
24 surviving children on mortality of widowed individuals.
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37 The following model can be used to estimate the association
38 between each of the five dummy variables and mortality:
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$$40 \quad h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

41 where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} ,
42 W_{65-74} , W_{75+} columns provided in the Table 2b.
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47 **Time since bereavement**

48 Next, we evaluated the association between mortality and time since bereavement
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9 or widowhood. We followed the approach of Schaefer et al. by considering the
10 following time since bereavement ranges: 0-6 months, 7-12 months, 13-24
11 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This
12 approach allowed us to estimate HR according to time since bereavement (see
13 Table 2c). The columns TSB₀₋₆, TSB₇₋₁₂, ... in Table 2c are time dependent
14 covariates that change with the survival time associated with widowed husbands
15 and wives. We did not account for remarriage in this analysis because of missing
16 remarriage dates.
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23 The following model was used to study the association between mortality and
24 time since bereavement:
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$$26 \quad h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

27
28 where W_1, \dots, W_7 dummy variables are associated with the TSB₀₋₆, TSB₇₋₁₂,
29 TSB₁₃₋₂₄, TSB₂₅₋₃₆, TSB₃₇₋₄₈, TSB₄₉₋₆₀ and TSB_{>60} columns provided in
30 the Table 2c.
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37 RESULTS

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39 We initially partitioned the 15,611 couples from AGDB into four cohorts
40 ranging in size from 3,210 (husband birth year 1850-1875) to 4,466 (husband
41 birth year 1876-1900). Wives were more likely to die after their husbands in all
42 cohorts, although in the three earliest cohorts (prior to 1850, 1850-1875, and
43 1876-1900), husbands had a higher mean age at death than their wives. The
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9 average age differences between the husbands and wives for all cohorts are
10 shown in Table 1. The number of remarried wives was far smaller than the
11 number of remarried husbands (n = 1,943 widowed husbands vs 613 widowed
12 wives). The number and proportion of remarried husbands and wives were
13 increased in the more contemporary cohorts (see Table 1). In contrast to other
14 populations from the 19th and early 20th centuries,⁴ the majority of widowed
15 husbands did not remarry, making it interesting to study the effect of remarriage
16 in the Amish population.
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23 **Hazard Ratios**

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25 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
26 associated with widowhood are displayed in Figure 42 according to birth cohort.
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28 These hazard ratios were estimated using the CPH model and design provided in
29 Table 2a. As expected, widowhood was associated with increased mortality for
30 both husbands and wives following the death of spouse, with HRs ranging
31 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
32 husbands in cohort 1850-1875). The impact of widowhood was
33 disproportionately greater on surviving husbands than on surviving wives in the
34 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
35 between widowed husbands vs wives in the 1850-1875 and 1901-1925 cohorts.
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37 There was no clear trend showing that the hazard ratios changed across cohorts
38 in either surviving husbands or wives. Since no clear differences were observed
39 between birth cohorts, all birth cohorts were combined to estimate the association
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9 between age at widowhood and mortality.

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11 Figure 23 shows the hazard ratios (and 95% CIs) for the model in which age
12 at widowhood is accounted for (design model in Table 2b). These results reveal
13 markedly higher bereavement effects in each age at widowhood group compared
14 to the results shown in Figure 42, consistent with the notion that the bereavement
15 effect may be diminished over time and most pronounced in the years proximal to
16 the death of the spouse.^{2 5 20 21} In nearly all age at widowhood categories, the
17 bereavement effect is stronger in widowed husbands than in widowed wives.
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24 Number of surviving children (> 6 vs ≤ 2) was included as a covariate in the
25 models whose results are shown in Figure 23. In general, there was a very weak
26 association between number of surviving children and mortality in widowed
27 husbands and widowed wives. Contrary to our expectations and a prior study,⁸ in
28 each case, a higher number of surviving children was associated with higher
29 mortality in the widowed husband/wife. When analyses were repeated without the
30 number of surviving children as a covariate, the estimates of the hazard ratios
31 were essentially unchanged (data not shown). Further, the results in Figure 23
32 show that the effect of bereavement decreases if the widowed individual
33 remarries.
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43 Hazard ratios and 95% CIs for the time since bereavement analysis are shown
44 in Figure 34. These results were obtained using the CPH model and the design
45 defined in Table 2c. The results show that there is an increase in risk of mortality
46 for recently widowed husbands and wives, and the hazard decreases with time
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9 since bereavement but remains significantly greater than 1. Further, the hazard is
10 higher (not significant) in wives vs husbands during the first 12 months following
11 bereavement. To address the issue that many individuals in the 1901-1925 cohort
12 may still have been alive at the last AGDB update and hence censored, we
13 performed the analysis for time since bereavement omitting the cohort 1901-1925.
14 The results show that the hazard ratios provided in Figure 4 have not changed
15 significantly; for example, for 25-26 months post bereavement the hazard ratios
16 change from 1.30 and 1.23 for all cohort to 1.25 and 1.17 for the three eldest
17 cohorts.

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26 Graphical checks of the overall adequacy of the CPH models were
27 performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a
28 reasonable fit to the data (data not shown). Further, ~~the~~ deviance residual plots
29 revealed ~~showed that there were~~ no obvious outliers in the data (data not shown).
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Further, the Wald test statistic was used to test the fit of the final model,¹⁸ and
according to this test statistic, the final model fits the data reasonably well
(Supplementary Tables S1, S2, and S3).

DISCUSSION

This is the first comprehensive study to evaluate the relationship between bereavement and social support in the Amish population. This study provides evidence that Amish widows and widowers have increased mortality risk

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9 compared to married cohort members. Although it is difficult to determine
10 whether this effect is of equivalent magnitude as that observed in studies of other
11 populations, the most recent studies of the bereavement effect using the CPH
12 suggest that our findings are generally consistent with data from other
13 populations.⁴

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18 Several previous studies on American, European, and Middle Eastern
19 populations have found that mortality is magnified in individuals widowed at a
20 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
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22 The LDS study is closest to our study because of the large family sizes and the
23 population selected as a religious isolate.⁸ The effects of bereavement on
24 mortality with respect to gender and the age at widowhood ranges observed in the
25 Amish are also largely consistent with those observed in the LDS population,⁸ and
26 other populations such as Finland and Israel (Figure 23).^{6 22} As expected,
27 widowers showed higher hazard ratios than widows except for the 1850-1875
28 cohort (Figure 12).

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37 In the present study, the association between bereavement and mortality is
38 greater in the first 6 months for both men and women (Figure 34), consistent with
39 previous findings.^{2 3 5 20 21 23} The mortality risks in the first 6 months are lower in
40 the Amish (Figure 34) compared to some studies,^{2 5 20 21} but not all studies.^{3 23} One
41 common pattern observed in this and other studies is that the initially high
42 bereavement effect first decreases but then increases with time since
43 bereavement.^{3 23} We speculate that the increased mortality during the first 6
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9 months might reflect acute effects related to the loss of a spouse, while the
10 gradual increases in mortality emerging in later life might reflect decreased
11 survival from aging-related diseases that is unmasked in the absence of spousal
12 support.
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16 Two strengths of our study are that, unlike many of the previously published
17 studies,^{5 13 16 20 21 24} ~~we did not need to incorporate censoring methods into our~~
18 ~~analysis because of the near completeness of birth and death dates of the Amish~~
19 ~~widows and widowers~~ we did not incorporate censoring methods into our analysis
20 because of the high availability of death dates of the Amish widows and widowers
21 in the first three cohorts; and we could study the effect of remarriage because
22 there were substantial numbers of widowed individuals who did and did not get
23 remarried (Table 1). Shor et al. noted that the difficulty in studying the effect of
24 remarriage in populations where all individuals are deceased because “In previous
25 decades, widowed men almost always remarried”.⁴ As suggested by previous
26 studies,^{8 9} remarriage after the death of a spouse significantly influences the
27 bereavement effect because it is associated with increased survival of both
28 widowers and widows across all cohorts and age at widowhood ranges. This
29 association is likely influenced by the fact that widowers or widows who get
30 remarried are in sufficiently good physical and mental health to do so. Further,
31 the survival rate is higher for widowers as compared to widows, possibly
32 reflecting support in Amish society for males getting remarried.
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48 Interestingly, increasing numbers of surviving children at the time of
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9 widowhood did not confer a survival advantage for Amish widows or widowers.
10 This result was counter to our hypothesis that children can help provide social
11 support for their parents. The hazard ratio was greater than 1.0 (but not
12 significant) for all widowers and widows with number of surviving children > 6
13 as compared to ≤ 2. Spouses in the Amish society may also provide unique
14 emotional, psychological, and social support to each other which cannot be
15 provided by their surviving children. The lack of protective association was
16 similarly observed when the number of surviving children was considered as a
17 linear or as a categorical variable (data not shown). This contrasts with data from
18 the Utah Population Database, in which increasing numbers of children were
19 associated with a decreased hazard ratio.⁸

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30 ~~Interestingly, increasing numbers of surviving children at the time of~~
31 ~~widowhood did not confer a survival advantage for Amish widows or widowers.~~
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33 ~~In fact, the hazard ratio was greater than 1.0 for all widowers and widows with~~
34 ~~number of surviving children > 6 as compared to ≤ 2. This result was counter to~~
35 ~~our hypothesis that children can help provide social support for their parents. The~~
36 ~~lack of protective association was similarly observed when the number of~~
37 ~~surviving children was considered as a linear or as a categorical variable (data not~~
38 ~~shown). This contrasts with data from the Utah Population Database, in which~~
39 ~~increasing numbers of children were associated with a decreased hazard ratio.⁸~~

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47 The implication of these findings is that differential familial support
48 following bereavement does not appear to be the key factor affecting mortality
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9 increases in widowed Amish populations. One potential explanation is that
10 spouses, as “attachment figures,” provide a unique, irreplaceable social support
11 which must be considered independently from other ancillary support providers,
12 such as children.²⁵ Data suggesting that relative mortality risk is also elevated in
13 divorced and never-married populations may support this hypothesis.³ In addition,
14 quality of social support has significantly greater effects on well-being than
15 quantity of support in elderly populations of both sexes.²⁶

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22 In adults aged 65 and older, lower reported social support has been associated
23 with decreased life satisfaction and increased depressive symptoms.²⁷ Family
24 members and close friends should be especially vigilant during the sensitive acute
25 period following the loss of a spouse, when relative mortality risk is the highest.

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32 A limitation of our data is that causes of death were unavailable. Future
33 research is needed to determine whether differential measures of social support
34 may be associated with certain causes of death in widowed populations. Divorce
35 in the Amish is sufficiently rare that this was not a major potential source of error.

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44 Our study results indicate that remarriage plays an important role in
45 improving the survival rate of widows and widowers in the Amish population.
46 Contrary to results from previous studies, an increase in the number of surviving
47 children was associated with a decreased survival rate of widowed individuals.

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Contributors BDM conceived the study. AS and SS did the research and data

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9 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
10 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
11 managed the project with assistance from BDM. AAS manages the AGDB project
12 at the National Institutes of Health and KAR managed the AGDB data at
13 University of Maryland Medical School during the period of this project. ARS
14 provided resources and advice. All authors proofread and approved the
15 manuscript.
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26 Geriatric Research and Education Center, Baltimore Veterans Administration
27 Medical Center.
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34 **Competing interests** None.
35
36

37 **Ethics approval** The construction and maintenance of AGDB are covered under
38 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
39 Investigator.
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43 **Provenance and peer review** Not commissioned; externally peer reviewed.
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46 **Data sharing statement** Data from the AGDB are available to investigators
47 (including ARS and various others not participating in this study) who have an
48 IRB-approved protocol to study the Amish or other Anabaptist groups.
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9 Investigators can request access to AGDB by writing to AAS and to Dr.
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11 Biesecker.

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FIGURE LEGENDS

Figure 1. A flow diagram which represents all the steps performed for filtering 15,611 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. "Unknown spouse" refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. "Birth year too late" means that the birth year of the husband is known and is > 1925. "Dates not recognized by R" are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. "Implausible birth or death dates" refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

Figure [42](#). Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a).

Figure [23](#). -Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).

Figure [34](#). -Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).

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TABLES

Table 1: Characteristics of 15,611 spouse pairs according to birth cohort of husband

	Cohorts			
	Pre-1850	1850-1875	1876-1900	1901-1925
Number of couples	3711	3210	4466	4224
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2360(53.0)	2439(57.7)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	2106(47.0)	1785(42.3)
Mean husband age at widowhood	63.0	59.8	62.2	67.9
Mean wife age at widowhood	62.0	62.8	66.5	69.6
Mean widowed husband survival in years	14.4	18.3	18.6	15.0
Mean widowed wife survival in years	15.3	16.0	17.0	15.1
Mean husband age at death	71.1	72.0	74.6	76.2
Mean wife age at death	69.6	68.5	72.7	77.0
Mean age difference husband-wife	3.4	2.8	2.1	1.3
Mean number of children	5.4	5.3	5.3	5.8
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	13(33.8)	588(33.0)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	221(9.4)	194(8.0)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

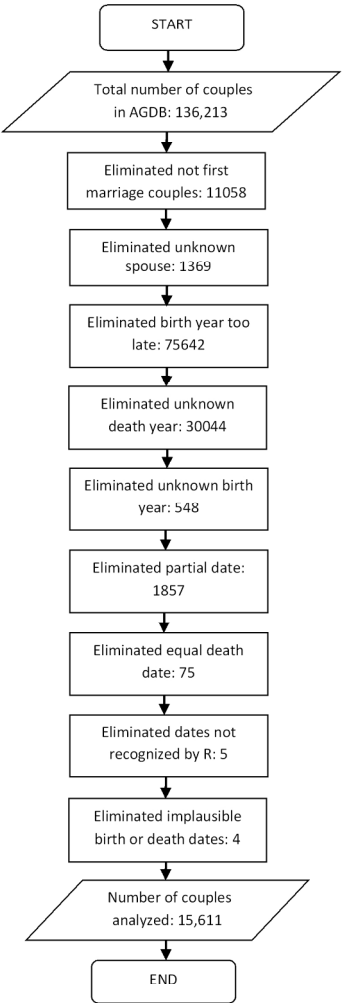
ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0

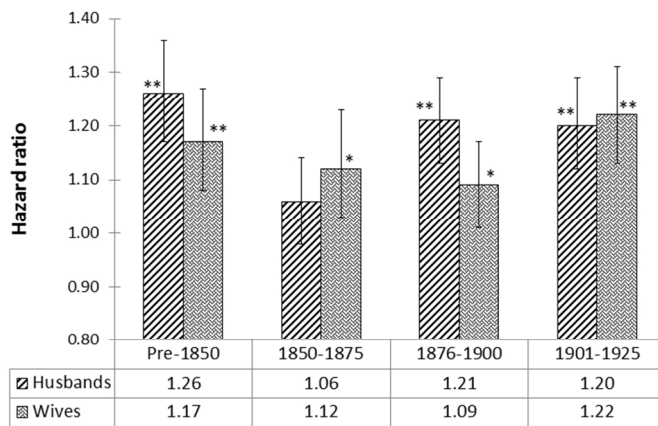
ID ₁	51	52	0	0	0	0	0	0	1	0
ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
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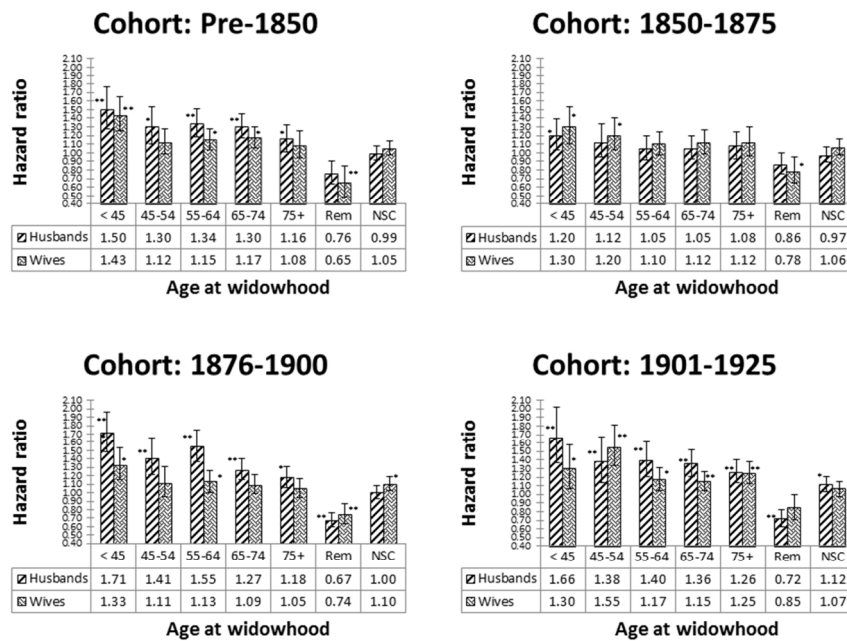


Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).
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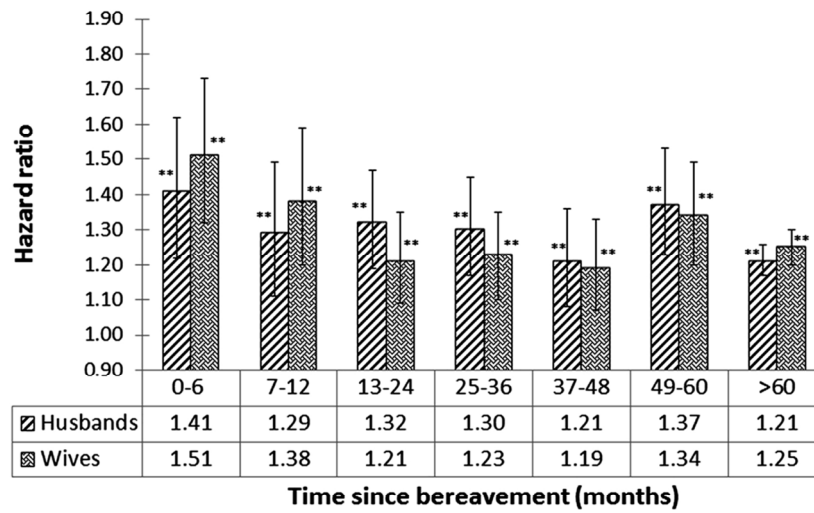


Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).
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Table S1: Wald statistics associated with models which are used to study the association between widowhood and mortality.

Cohorts				
	Pre-1850	1850-1875	1876-1900	1901-1925
Husbands	38.2 ^{**}	5.9 [*]	33.8 ^{**}	28.9 ^{**}
Wives	15.2 ^{**}	6.9 [*]	4.8 [*]	26.9 ^{**}
* P<0.05 and ** P<0.001				

Table S2: Wald statistics associated with models which are used to study the association between timing of widowhood and mortality.

Cohorts				
	Pre-1850	1850-1875	1876-1900	1901-1925
Husbands	57.2 ^{**}	15.8 [*]	90.6 ^{**}	64.4 ^{**}
Wives	39.8 ^{**}	20.3 ^{**}	30.4 ^{**}	50.5 ^{**}
* P<0.05 and ** P<0.001				

Table S3: Wald statistics associated with models which are used to study the association between time since bereavement and mortality.

Husbands	154.3 ^{**}
Wives	147.8 ^{**}
** P<0.001	

STROBE Statement—Checklist of items that should be included in reports of *cohort studies*

	Item No	Recommendation
Title and abstract	1	<p>(a) Indicate the study's design with a commonly used term in the title or the abstract (From Abstract) This study investigates the association between bereavement and the mortality of a surviving spouse among 15,611 Amish couples born during 1725-1925. We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using the Cox proportional hazard model.</p> <p>(b) Provide in the abstract an informative and balanced summary of what was done and what was found. See Abstract for findings.</p>
Introduction		
Background/rationale	2	<p>Explain the scientific background and rationale for the investigation being reported (From Introduction) It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the "bereavement effect," is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.²⁻⁵ Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.⁶⁻⁷ To gain a different perspective on potential influences of social support on bereavement, we assessed the bereavement effect in an Amish population, whose culture is characterized by its core beliefs in community and social cohesion. Examples of the strong degree of social support within the Amish include community-managed health insurance and community ties through membership in the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹² We reasoned that the high level of social support in the Amish population might mitigate the bereavement effect.</p>
Objectives	3	<p>State specific objectives, including any prespecified hypotheses (From Abstract) We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support. We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using the Cox proportional hazard model.</p>
Methods		
Study design	4	<p>Present key elements of study design early in the paper (From Introduction) We used Cox proportional hazard (CPH) models to analyze the association of bereavement and mortality of widowed husbands and wives. We considered remarriage and number of surviving children as additional potential modifiers of the bereavement effect. (From Material and Methods). The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement of the Amish population.¹⁴⁻¹⁵</p>
Setting	5	<p>Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection (From Material and Methods) The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement on the Amish population.¹⁴⁻¹⁵ The AGDB is a computerized genealogy database of Amish and some Mennonite populations, last updated in 2010. The AGDB contains information for 539,822 Amish individuals and 136,213 Amish couples. This database includes information about the family relationships and the birth, marriage, and death dates of children of Amish individuals</p>

		from North America, mostly located in Pennsylvania, Ohio, and Indiana.
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up (From Materials and Methods) For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1926; (2) their birth and death dates were recorded; (3) they were in their first marriage; and (4) they did not die on the same day. Constraint (4) excludes couples who may have died from a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 31, 1925. Our data set included 15,611 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood.
		(b) For matched studies, give matching criteria and number of exposed and unexposed This is not a matched study
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable See Material and Methods, especially Table 2.
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group (From Material and Methods) The Anabaptist Genealogy Database (AGDB) version 5 was used to study the effect of bereavement on the Amish population. ^{14 15} The AGDB is a computerized genealogy database of Amish and some Mennonite populations, last updated in 2010. The AGDB contains information for 539,822 Amish individuals and 136,213 Amish couples. This database includes information about the family relationships and the birth, marriage, and death dates of children of Amish individuals from North America, mostly located in Pennsylvania, Ohio, and Indiana.
Bias	9	Describe any efforts to address potential sources of bias We excluded couples who died on the same day. The data are not censored.
Study size	10	Explain how the study size was arrived at All couples in AGDB meeting the criteria described in item 5.
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why (From Material and Methods) For some analyses, these couples were first partitioned into eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-1900, and 1901-1925) and sex. For the time since bereavement analyses, we did not partition into cohorts because the number of widow(er)s who died within 6 months after being bereaved is small (N=224). Characteristics of these couples by husband birth cohort are shown in Table 1.
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding See Material and Methods, especially Table 2. (b) Describe any methods used to examine subgroups and interactions (From Material and Methods) For some analyses, these couples were first partitioned into eight cohorts based on the husband's birth year (prior to 1850, 1850-1875, 1876-1900, and 1901-1925) and sex.

		(c)	Explain how missing data were addressed Couples with missing dates were excluded.
		(d)	If applicable, explain how loss to follow-up was addressed Not applicable.
		(e)	Describe any sensitivity analyses Not applicable
Results			
Participants	13*	(a)	Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed See Table 1
		(b)	Give reasons for non-participation at each stage Not applicable
		(c)	Consider use of a flow diagram We preferred to describe the analysis designs in Table 2.
Descriptive data	14*	(a)	Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders See Table 1.
		(b)	Indicate number of participants with missing data for each variable of interest Couples with missing dates were excluded.
		(c)	Summarise follow-up time (eg, average and total amount) This is a retrospective study; All participants are deceased.
Outcome data	15*		Report numbers of outcome events or summary measures over time See Table 1.
Main results	16	(a)	Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included See Figures 1-3 and text of Results. Figures do show 95% confidence intervals.
		(b)	Report category boundaries when continuous variables were categorized See Table 2.
		(c)	If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period Not applicable.
Other analyses	17		Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses See Results text.

Discussion		
Key results	18	<p>Summarise key results with reference to study objectives</p> <p>(From Discussion) This study provides evidence that Amish widows and widowers have increased mortality risk compared to married cohort members. Although it is difficult to determine whether this effect is of equivalent magnitude as that observed in studies of other populations, the most recent studies of the bereavement effect using the CPH suggest that our findings are generally consistent with data from other populations.⁴</p> <p>Interestingly, increasing numbers of surviving children at the time of widowhood did not confer a survival advantage for Amish widows or widowers. In fact, the hazard ratio was greater than 1.0 for all widowers and widows with number of surviving children > 6 as compared to ≤ 2. This result was counter to our hypothesis that children can help provide social support for their parents.</p>
Limitations	19	<p>Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias</p> <p>(From Discussion) A limitation of our data is that causes of death were unavailable. Future research is needed to determine whether differential measures of social support may be associated with certain causes of death in widowed populations. Divorce in the Amish is sufficiently rare that this was not a major potential source of error.</p>
Interpretation	20	<p>Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence</p> <p>(From Discussion) The effects of bereavement on mortality with respect to gender and the age at widowhood ranges observed in the Amish are also largely consistent with those observed in the LDS population,⁸ and other populations such as Finland and Israel (Figure 2).^{6,22} As expected, widowers showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure 1).</p> <p>Two strengths of our study are that, unlike many of the previously published studies,^{5,13,16,20,21,24} we did not need to incorporate censoring methods into our analysis because of the near completeness of birth and death dates of the Amish widows and widowers; and we could study the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1).</p> <p>Our study results indicate that remarriage plays an important role in improving the survival rate of widows and widowers in the Amish population. Contrary to results from previous studies, an increase in the number of surviving children was associated with a decreased survival rate of widowed individuals.</p>
Generalisability	21	<p>Discuss the generalisability (external validity) of the study results</p> <p>(From Discussion) One common pattern observed in this and other studies is that the initially high bereavement effect first decreases but then increases with time since bereavement.^{3,23} We speculate that the increased mortality during the first 6 months might reflect acute effects related to the loss of a spouse, while the gradual increases in mortality emerging in later life might reflect decreased survival from aging-related diseases that is unmasked in the absence of spousal support.</p> <p>As suggested by previous studies,^{8,9} remarriage after the death of a spouse significantly influences the bereavement effect because it is associated with increased survival of both widowers and widows across all cohorts and age at widowhood ranges. This association is likely influenced by the fact that widowers or widows who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for widowers as compared to widows, possibly reflecting support in Amish society for males getting remarried.</p>

1 Our study results indicate that remarriage plays an important role in improving the
2 survival rate of widows and widowers in the Amish population.
3

Other information

4
5 Funding 22 Give the source of funding and the role of the funders for the present study and, if
6 applicable, for the original study on which the present article is based
7 (From Acknowledgments) This work was supported by research grants R01 HL69313,
8 R01 DK54261, R01 AG1872801, R01 HL088119, R01 AR046838, and U01 HL72515
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10 Intramural Research Program of the National Institutes of Health, NLM and the
11 Geriatric Research and Education Center, Baltimore Veterans Administration Medical
12 Center. These funders had no role in the study itself.
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16 *Give information separately for exposed and unexposed groups.
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18 **Note:** An Explanation and Elaboration article discusses each checklist item and gives methodological background and
19 published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely
20 available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at
21 <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is
22 available at <http://www.strobe-statement.org>.
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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Abstract: 239

Introduction through Discussion: 2,988.

ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 10,892 Amish couples born during 1725-1900 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure The survival time is 'age'; event is 'death'. Hazard ratios of widowed individuals with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both males and females. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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3 socially cohesive Amish community. Remarriage is associated with a significant
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5 decrease in the mortality risk among Amish individuals. Contrary to results from
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7 previous studies, an increase in the number of surviving children was associated
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9 with decreased survival rate.
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For peer review only

ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.
- We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks among Amish individuals, and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.
- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.

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- The number of surviving children was simplified to be a time independent covariate although it may change between the death of the first parent and the death of the second parent.

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INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.^{2 5} Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6 7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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The Amish maintain a cultural identity distinct from mainstream American

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3 culture that is characterized by their traditional dress, a plain lifestyle, and non-
4 adoption of modern technology (e.g., electricity, cars, telephones), German
5 dialect, separate school system, and ultra-conservative Anabaptist religious
6 practices. A central tenet of Amish culture throughout their history in the USA
7 has been social cohesiveness with emphasis on family and community. Members
8 of this tight-knit society have extraordinary social support from cradle to grave,
9 including community-financing of medical costs and support during times of
10 need. The community financing of medical costs became increasingly formalized
11 during the 20th century to the point where it is now a formal system of self-
12 insurance called “Amish Aid”.
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27 To gain a different perspective on potential influences of social support on
28 bereavement, we assessed the bereavement effect in an Amish population, whose
29 culture is characterized by its core beliefs in community and social cohesion.
30 Examples of the strong degree of social support within the Amish include
31 community-managed health insurance and community ties through membership in
32 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
33 We reasoned that the high level of social support in the Amish population might
34 mitigate the bereavement effect. Previous studies used covariates such as
35 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
36 neighborhood interaction, and household size to study the relationship between
37 bereavement and social support.^{4 8 13}
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53 We estimated bereavement effects in widows and widowers separately, and
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3 further assessed mortality risks as a function of age at widowhood and time since
4 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
5 association of bereavement and mortality of widowed husbands and wives. We
6 considered remarriage and number of surviving children as additional potential
7 modifiers of the bereavement effect.
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14 15 16 17 **MATERIAL AND METHODS**

18 19 **Data source**

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21 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
22 effect of bereavement on the Amish population.^{14 15} The AGDB is a
23 computerized genealogy database of Amish and some Mennonite populations, last
24 updated in 2010. This database includes information about the family
25 relationships and the birth, marriage, and death dates of children of Amish
26 individuals from North America, mostly located in Pennsylvania, Ohio, and
27 Indiana. The "individual table" of AGDB contains information about 539,822
28 individuals. The "relationship table" includes information about 136,213 Amish
29 couples. An individual who is married multiple times participates in multiple
30 relationship table entries. There are 1,369 relationship entries among the 136,213
31 entries concerning children for whom one or both biological parents are unknown
32 (Figure 1).
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50 Amish people rarely give birth out of wedlock, and a vast majority of the
51 mating samples we used are documented as marriages in the three original sources
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3 from which AGDB is derived. However, there is less availability of marriage
4 dates than birth dates; we did not wish to exclude couples with missing marriage
5 dates. For this study, we included couples in which both spouses met four
6 conditions: (1) they were born prior to January 1, 1901; (2) their birth and death
7 dates were recorded; (3) they were in their first marriage; and (4) they did not die
8 on the same day. Constraint (4) excludes couples who may have died from a
9 shared disaster (e.g., accident). A total of 10,892 couples met the inclusion
10 criteria. The oldest person included was born on February 14, 1725, and the
11 youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were
12 applied.
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27 Our data set included 10,892 couples with information on their date of birth,
28 date of death, number of surviving children, family IDs, and remarriage status
29 after widowhood. For some analyses, these couples were first partitioned into
30 three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and
31 based on both husband's and wife's birth year (1876-1900) and sex. For the time
32 since bereavement analyses, we did not partition into cohorts because the number
33 of widow(er)s who died within 6 months after being bereaved is small (N=291).
34 Characteristics of these couples are shown in Table 1.
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47 **Statistical analyses**

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49 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
50 Hazards (CPH) model where the response variable or survival time is 'age at
51 death' and the event is 'death'. The model is used to study the association of
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3 widowhood and mortality rates in the surviving spouse, while adjusting for
4 covariates such as education, health habits, age in years, number of children,
5 and remarriage. In some of our analyses, we adjusted for remarriage and
6 number of children as covariates; we did not adjust for education or health
7 habits. Husbands and wives were always analyzed separately.

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15 The main covariate, widowhood, was monitored as a time-dependent
16 covariate, by assigning a value of 1 for each time period the individual was
17 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
18 covariate, by assigning a value 1 for the widowed period if the individual
19 remarried and 0 otherwise. The number of surviving children was a time-
20 independent covariate and counted at the beginning of widowhood period of the
21 surviving spouse. The covariate was divided into three categories, as follows:
22 0=number of surviving children is ≤ 2 , 1=number of surviving children is between
23 3 and 6, and 2=number of surviving children is > 6 . The categories ≤ 2 children, 3-
24 6 children, and > 6 children are separate. These boundaries were chosen to ensure
25 categories that were roughly balanced in size. To evaluate any possible bias by
26 counting the number of surviving children at the death of the first spouse, we also
27 estimated how many couples changed categories between the death of the first
28 spouse and the death of second spouse due to death of one or more children
29 during the widowhood period.

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51 Survival analyses were performed using the R programming environment.¹⁷
52
53 All estimates and confidence intervals were obtained using the `coxph` function
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3 available in the `survival` package.
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6 **Representation of survival data for CPH analysis**

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8 We show below examples of how we represented the survival data for CPH
9 analysis using a time modeling approach. To represent the survival data columns, the
10 standard approach is to convert the couple's demographic information, date of
11 death, date of birth, remarriage and number of surviving children into columns
12 representing **start** time, **stop** time, **event** status, and all of the included covariates
13 (widowhood, remarriage, and number of surviving children).^{18 19} Here (**start**,
14 **stop**] is an interval of risk, open on the left and closed on the right; the **event**
15 column is set to 1 if the subject had an event (death) at time **stop** and is 0
16 otherwise. To illustrate the subtleties, we consider a hypothetical example of
17 three wives (ID1, ID2 and ID3) from the cohort 1850-1875.
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- 34 • *ID*1: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
35 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
36
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- 40 • *ID*2: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
41 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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- 48 • *ID*3: born on 03/01/1871; never widowed and never remarried; number of
49 surviving children = 4 and died on 04/01/1930 at age 59.
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3 The data used for the CPH considering widowhood as a single time-
4 dependent covariate is presented in Table 2a. The following model was used to
5 estimate the association between widowhood and mortality.
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$$10 \quad h(t) = h_0(t) \exp[\beta W]$$

11
12 This model can be run with additional covariates and results that adjust for the
13 ability to remarry and the number of surviving children are presented.
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17 **Timing of widowhood**

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19 The above model considers widowhood as a single time-dependent
20 covariate, evaluating the association between mortality and being
21 widowed. One can further ask if that association varies over the life
22 course by considering the impact of age at widowhood on mortality.
23
24 We addressed this issue following the approach of Mineau et al. by
25 adding terms for age at widowhood into the model as covariates (<45,
26 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to
27 age at widowhood (see Table 2b).⁸ These terms were included in the model
28 as time-dependent dummy variables, created to represent the
29 widowhood experience of each individual across the 5 age windows
30 spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} .
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32 Further, we also addressed the impact of remarriage and number of
33 surviving children on the mortality of Amish individuals.
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51 The following model can be used to estimate the association
52 between each of the five dummy variables and mortality:
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$$h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , W_{75+} columns provided in the Table 2b.

Time since bereavement

Next, we evaluated the association between mortality and time since bereavement or widowhood. We followed the approach of Schaefer et al. by considering the following time since bereavement ranges: 0-6 months, 7-12 months, 13-24 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This approach allowed us to estimate HR according to time since bereavement (see Table 2c). The columns TSB_{0-6} , TSB_{7-12} , ... in Table 2c are time dependent covariates that change with the survival time associated with widowed husbands and wives. We did not account for remarriage in this analysis because of missing remarriage dates.

The following model was used to study the association between mortality and time since bereavement:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

where W_1, \dots, W_7 dummy variables are associated with the TSB_{0-6} , TSB_{7-12} , TSB_{13-24} , TSB_{25-36} , TSB_{37-48} , TSB_{49-60} and $TSB_{>60}$ columns provided in the Table 2c.

RESULTS

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2
3 We initially partitioned the 10,892 couples from AGDB into three cohorts ranging
4 in size from 3,210 (husband birth year 1850-1875) to 3,971 (husband birth year
5 1876-1900). Wives were more likely to die after their husbands in all three
6 cohorts while husbands had a higher mean age at death than their wives. The
7 average age differences between the husbands and wives for all cohorts are shown
8 in Table 1. The number of remarried wives was far smaller than the number of
9 remarried husbands (n = 1,290 widowed husbands vs 378 widowed wives). The
10 number and proportion of remarried husbands and wives were increased in the
11 more contemporary cohorts (see Table 1). In contrast to other populations from
12 the 19th and early 20th centuries,⁴ the majority of widowed husbands did not
13 remarry, making it interesting to study the effect of remarriage in the Amish
14 population.

31 Hazard Ratios

32 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
33 associated with widowhood are displayed in Figure 2 according to birth cohort.
34 The significance of these hazard ratios are indicated with the help of p-values
35 (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2.
36 These hazard ratios were estimated using the CPH model and design provided in
37 Table 2a. As expected, widowhood was associated with increased mortality for
38 both husbands and wives following the death of spouse, with HRs ranging
39 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
40 husbands in cohort 1850-1875). The impact of widowhood was
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3 disproportionately greater on surviving husbands than on surviving wives in the
4 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
5 between widowed husbands vs. wives in the 1850-1875. There was no clear trend
6 showing that the hazard ratios changed across cohorts in either surviving
7 husbands or wives.
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Figure 3 shows the hazard ratios along with 95% CIs, and range of p-values for the model in which age at widowhood is accounted for (design model in Table 2b). These results reveal markedly higher bereavement effects in each age at widowhood group compared to the results shown in Figure 2, consistent with the notion that the bereavement effect may be diminished over time and most pronounced in the years proximal to the death of the spouse.^{2 5 20 21} In nearly all age at widowhood categories, the bereavement effect is stronger in widowed husbands than in widowed wives.

The number of children surviving when the first spouse died (with two pairwise comparisons 3-6 vs ≤ 2 ; > 6 vs ≤ 2) was included as a time-independent covariate in the models whose results are shown in Figure 3. In general, there was a very weak association between number of surviving children and mortality in husbands and wives. Contrary to our expectations and a prior study,⁸ in each case, a higher number of surviving children was not significantly associated with lower mortality in both husbands and wives (See Figure 3). Further, the results in Figure 3 show that the effect of bereavement decreases if the Amish individual remarries.

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There are two potential sources of bias in analyses involving the number of surviving children covariate. Couples in which one spouse died at age <50 are likely not to have as many children as couples in which both parents survived to at least age 50. To quantify this bias, we repeated the analysis in Figure 3 by excluding all the couples who got widowed before age 50 (data not shown). The results show that there is no significant change in the hazard ratios vs. Figure 3. The maximum change for any of the hazard ratios related to the number of surviving children was 0.03 (data not shown).

On the other end of the age spectrum, there is a potential source of bias as more children may die, the longer the surviving parent lives. Because the number of children was divided into three categories ≤ 2 , 3-6, and > 6 , any possible bias could arise when and only when the death of a child shifted the number of surviving children from a higher category to a lower category. Over all couples, the number of widows or widowers whose number of surviving children changed to a lower category from the date of widowhood to the date of death was only 298 (< 3% of all couples).

When the analyses corresponding to Figure 3 were repeated excluding these 298 couples, the hazard ratios were essentially unchanged. The maximum change was 0.04; sometimes the hazard ratio increased slightly and sometime it decreased slightly (data not shown).

Hazard ratios and 95% CIs for the time since bereavement analysis are shown in Figure 4. The significant p-values are indicated in Figure 4. These results were

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obtained using the CPH model and the design defined in Table 2c. The results show that there is an increase in risk of mortality for recently widowed husbands and wives, and the hazard decreases with time since bereavement but remains significantly greater than 1. Further, the hazard is higher (not significant) in wives vs. husbands during the first 12 months following bereavement.

The range of p-values provided in Figures 2-4 indicates the significance of each of the hazard ratios. For example, in Figure 4, the p-value = 0.0001 associated with the hazard ratio = 1.39 (males; time since bereavement range < 6 months) strongly indicates that the hazard ratio is significantly > 1. Similarly, the p-value = 0.039 associated with the hazard ratio = 1.15 (males; time since bereavement 37-48 months) weakly indicates the significance of the hazard ratio > 1.

Graphical checks of the overall adequacy of the CPH models were performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a reasonable fit to the data (data not shown). The *deviance residual* plots revealed no obvious outliers in the data (data not shown). Further, the Wald test statistic was used to test the fit of the final model,¹⁸ and according to this test statistic, the final model fits the data reasonably well (Supplementary Tables S1, S2, and S3).

DISCUSSION

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3 This is the first comprehensive study to evaluate the relationship between
4 bereavement and social support in the Amish population. This study provides
5 evidence that Amish widows and widowers have increased mortality risk
6 compared to married cohort members. Although it is difficult to determine
7 whether this effect is of equivalent magnitude as that observed in studies of other
8 populations, the most recent studies of the bereavement effect using the CPH
9 suggest that our findings are generally consistent with data from other
10 populations.⁴

11
12 Several previous studies on American, European, and Middle Eastern
13 populations have found that mortality is magnified in individuals widowed at a
14 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
15 The LDS study is closest to our study because of the large family sizes and the
16 population selected as a religious isolate.⁸ The effects of bereavement on
17 mortality with respect to gender and the age at widowhood ranges observed in the
18 Amish are also largely consistent with those observed in the LDS population,⁸ and
19 other populations such as Finland and Israel (Figure 3).^{6 22} As expected, widowers
20 showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure
21 2).

22 In the present study, the association between bereavement and mortality is
23 greater in the first 6 months for both men and women (Figure 4), consistent with
24 previous findings.^{2 3 5 20 21 23} The mortality risks in the first 6 months are lower in
25 the Amish (Figure 4) compared to some studies,^{2 5 20 21} but not all studies.^{3 23} One

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4 common pattern observed in this and other studies is that the initially high
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6 bereavement effect first decreases but then increases with time since
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8 bereavement.^{3 23} We speculate that the increased mortality during the first 6
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10 months might reflect acute effects related to the loss of a spouse, while the
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12 gradual increases in mortality emerging in later life might reflect decreased
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14 survival from aging-related diseases that is unmasked in the absence of spousal
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16 support.
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20 One strength of our study is that we could evaluate the effect of remarriage
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22 because there were substantial numbers of widowed individuals who did and did
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24 not get remarried (Table 1) Shor et al. noted that the difficulty in studying the
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26 effect of remarriage in populations where all individuals are deceased because “In
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28 previous decades, widowed men almost always remarried”.⁴ As suggested by
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30 previous studies,^{8 9} remarriage after the death of a spouse significantly influences
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32 the bereavement effect because it is associated with increased survival of both
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34 males and females across all cohorts . This association is likely influenced by the
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36 fact that males or females who get remarried are in sufficiently good physical and
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38 mental health to do so. Further, the survival rate is higher for males as compared
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40 to females, possibly reflecting support in Amish society for males getting
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42 remarried.
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48 Interestingly, increasing numbers of surviving children at the time of
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50 widowhood did not confer a survival advantage for Amish individuals. This result
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52 was counter to our hypothesis that children can help provide social support for
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3 their parents. The hazard ratio was greater than 1.0 (but not significant) for all
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5 Amish individuals with number of surviving children > 6 as compared to ≤ 2 .
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8 Spouses in the Amish society may also provide unique emotional, psychological,
9
10 and social support to each other which cannot be provided by their surviving
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12 children. The lack of protective association was similarly observed when the
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14 number of surviving children was considered as a linear or as a categorical
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16 variable (data not shown). This contrasts with data from the Utah Population
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18 Database, in which increasing numbers of children were associated with a
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20 decreased hazard ratio.⁸
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25 The implication of these findings is that differential familial support
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27 following bereavement does not appear to be the key factor affecting mortality
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29 increases in widowed Amish populations. One potential explanation is that
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31 spouses, as “attachment figures,” provide a unique, irreplaceable social support
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33 which must be considered independently from other ancillary support providers,
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35 such as children.²⁴ Data suggesting that relative mortality risk is also elevated in
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37 divorced and never-married populations may support this hypothesis.³ In addition,
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39 quality of social support has significantly greater effects on well-being than
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41 quantity of support in elderly populations of both sexes.²⁵
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46 In adults aged 65 and older, lower reported social support has been associated
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48 with decreased life satisfaction and increased depressive symptoms.²⁶ Family
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50 members and close friends should be especially vigilant during the sensitive acute
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52 period following the loss of a spouse, when relative mortality risk is the highest.
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There are two limitations of our data. First, the number of surviving children covariate was simplified because it was treated as a time-independent covariate, although some children die before their parents, so it could be considered as time-dependent instead. Second, the causes of death were unavailable. Future research is needed to study the effect of the loss of a child on the mortality of husbands and wives by considering the number of surviving children as a time-dependent covariate. Also, research is needed to determine whether differential measures of social support may be associated with certain causes of death in widowed populations. Divorce in the Amish is sufficiently rare that this was not a major potential source of error.

Our study results indicate that remarriage plays an important role in improving the survival rate of males and females in the Amish population. Contrary to results from previous studies, an increase in the number of surviving children was associated with a decreased survival rate of Amish individuals.

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3 provided resources and advice. All authors proofread and approved the
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5 manuscript.
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15
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18 Medical Center.
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23 **Competing interests** None.
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26 **Ethics approval** The construction and maintenance of AGDB are covered under
27
28 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
29
30 Investigator.
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34 **Provenance and peer review** Not commissioned; externally peer reviewed.
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37 **Data sharing statement** Data from the AGDB are available to investigators
38
39 (including ARS and various others not participating in this study) who have an
40
41 IRB-approved protocol to study the Amish or other Anabaptist groups.
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43 Investigators can request access to AGDB by writing to AAS and to Dr.
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FIGURE LEGENDS

Figure 1. A flow diagram that represents all the steps performed for filtering 10,892 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. “Unknown spouse” refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. “Birth year too late” means that the birth year of the husband or wife is known and is > 1901 . “Dates not recognized by R” are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. “Implausible birth or death dates” refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a); * and ** on the top of the blocks represent significance of hazard ratios with $p\text{-value} < 0.05$ and $p\text{-value} < 0.001$, respectively.

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood (design provided in Table 2b); NSC1: Number of Surviving Children (3-6 vs ≤ 2); NSC2: Number of Surviving

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3 Children (>6 vs ≤ 2); * and ** on the top of the blocks represent significance of
4 hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.
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8 Figure 4. Hazard ratios of widowed husbands and wives versus their married
9 counterparts according to time since bereavement (months) (design provided in
10 Table 2c); * and ** on the top of the blocks represent significance of hazard ratios
11 with p-value < 0.05 and p-value < 0.001, respectively.
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TABLES

Table 1: Characteristics of 10,892 spouse pairs according to birth cohort of husband.

Cohorts	Pre-1850	1850-1875	1876-1900
Number of couples	3711	3210	3971
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2043(51.4)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	1928(48.2)
Mean husband age at widowhood	63.0	59.8	62.2
Mean wife age at widowhood	62.0	62.8	66.9
Mean widowed husband survival in years	14.4	18.3	18.4
Mean widowed wife survival in years	15.3	16.0	16.6
Mean husband age at death	71.1	72.0	74.6
Mean wife age at death	69.6	68.5	72.6
Mean age difference husband-wife	3.4	2.8	2.0
Mean number of children	5.4	5.3	5.3
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	648(33.6)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	180(8.8)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0
ID ₁	51	52	0	0	0	0	0	0	1	0

ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Abstract: 239

Introduction through Discussion: 2,988.

ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

~~Participants 10,892 Amish couples born during 1725-1900 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.~~

~~Participants 15,611 Amish couples born during 1725-1925 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.~~

Outcome measure The survival time is 'age'; event is 'death'. Hazard ratios of widowed individuals with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both males and females. ~~We observed a higher number of surviving children to be associated with~~

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9 ~~increased mortality in both widows and widowers.~~ Mortality risk following
10 bereavement was higher in the first 6 months among both men and women.

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12 **Conclusions** We conclude that bereavement effects remain apparent even in this
13 socially cohesive Amish community. Remarriage is associated with a significant
14 decrease in the mortality risk among Amish individuals. ~~Remarriage is associated~~
15 ~~with a significant decrease in the mortality risk among widowed individuals.~~
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20 Contrary to results from previous studies, an increase in the number of surviving
21 children was associated with decreased survival rate.
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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.
- We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks among Amish individuals, and therefore the support provided by the spouse is irreplaceable.~~This study shows that strong social support is not sufficient to decrease the mortality risks of the bereaved individuals and therefore the support provided by the spouse is irreplaceable.~~

- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- ~~Due to the availability of a large and uncensored dataset, our hazard ratio estimates are mostly significant with narrow confidence intervals.~~
- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.
- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.
- The number of surviving children was simplified to be a time independent covariate although it may change between the death of the first parent and the death of the second parent.

INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.²⁻⁵ Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6,7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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The Amish maintain a cultural identity distinct from mainstream American

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9 culture that is characterized by their traditional dress, a plain lifestyle, and non-
10 adoption of modern technology (e.g., electricity, cars, telephones), German
11 dialect, separate school system, and ultra-conservative Anabaptist religious
12 practices. A central tenet of Amish culture throughout their history in the USA
13 has been social cohesiveness with emphasis on family and community. Members
14 of this tight-knit society have extraordinary social support from cradle to grave,
15 including community-financing of medical costs and support during times of
16 need. The community financing of medical costs became increasingly formalized
17 during the 20th century to the point where it is now a formal system of self-
18 insurance called “Amish Aid”.~~Members of this tight knit society have~~
19 ~~extraordinary social support from cradle to grave, including community managed~~
20 ~~health insurance and support during times of need. Elderly parents are taken care~~
21 ~~of by their children and neighbors; they do not use assisted living or nursing~~
22 ~~homes to care for their elderly.~~
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35 To gain a different perspective on potential influences of social support on
36 bereavement, we assessed the bereavement effect in an Amish population, whose
37 culture is characterized by its core beliefs in community and social cohesion.
38 Examples of the strong degree of social support within the Amish include
39 community-managed health insurance and community ties through membership in
40 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
41 We reasoned that the high level of social support in the Amish population might
42 mitigate the bereavement effect. Previous studies used covariates such as
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9 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
10 neighborhood interaction, and household size to study the relationship between
11 bereavement and social support.^{4 8 13}

14 We estimated bereavement effects in widows and widowers separately, and
15 further assessed mortality risks as a function of age at widowhood and time since
16 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
17 association of bereavement and mortality of widowed husbands and wives. We
18 considered remarriage and number of surviving children as additional potential
19 modifiers of the bereavement effect.
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27 MATERIAL AND METHODS

29 Data source

30 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
31 effect of bereavement on the Amish population.^{14 15} The AGDB is a
32 computerized genealogy database of Amish and some Mennonite populations, last
33 updated in 2010. This database includes information about the family
34 relationships and the birth, marriage, and death dates of children of Amish
35 individuals from North America, mostly located in Pennsylvania, Ohio, and
36 Indiana. The "individual table" of AGDB contains information about 539,822
37 individuals. The "relationship table" includes information about 136,213 Amish
38 couples. An individual who is married multiple times participates in multiple
39 relationship table entries. There are 1,369 relationship entries among the 136,213
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9 entries concerning children for whom one or both biological parents are unknown
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11 (Figure 1).

12 Amish people rarely give birth out of wedlock, and a vast majority of the
13 mating samples we used are documented as marriages in the three original sources
14 from which AGDB is derived. However, there is less availability of marriage
15 dates than birth dates; we did not wish to exclude couples with missing marriage
16 dates. For this study, we included couples in which both spouses met four
17 conditions: (1) they were born prior to January 1, 1901; (2) their birth and death
18 dates were recorded; (3) they were in their first marriage; and (4) they did not die
19 on the same day. Constraint (4) excludes couples who may have died from a
20 shared disaster (e.g., accident). A total of 10,892 couples met the inclusion
21 criteria. The oldest person included was born on February 14, 1725, and the
22 youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were
23 applied.~~Amish people rarely give birth out of wedlock, and a vast majority of the~~
24 ~~mating samples we used are documented as marriages in the three original sources~~
25 ~~from which AGDB is derived. However, there is less availability of marriage~~
26 ~~dates than birth dates; we did not wish to exclude couples with missing marriage~~
27 ~~dates. For this study, we included couples in which both spouses met four~~
28 ~~conditions: (1) they were born prior to January 1, 1926; (2) their birth and~~
29 ~~death dates were recorded; (3) they were in their first marriage; and (4) they did~~
30 ~~not die on the same day. Constraint (4) excludes couples who may have died from~~
31 ~~a shared disaster (e.g., accident). A total of 15,611 couples met the inclusion~~
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9 ~~criteria. The oldest person included was born on February 14, 1725, and the~~
10 ~~youngest on December 31, 1925. See Figure 1 for how the exclusion criteria were~~
11 ~~applied.~~

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14 Our data set included 10,892 couples with information on their date of birth,
15 date of death, number of surviving children, family IDs, and remarriage status
16 after widowhood. For some analyses, these couples were first partitioned into
17 three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and
18 based on both husband's and wife's birth year (1876-1900) and sex. For the time
19 since bereavement analyses, we did not partition into cohorts because the number
20 of widow(er)s who died within 6 months after being bereaved is small (N=291).
21 Characteristics of these couples are shown in Table 1.~~Our data set included 15,611~~
22 ~~couples with information on their date of birth, date of death, number of surviving~~
23 ~~children, family IDs, and remarriage status after widowhood. For some analyses,~~
24 ~~these couples were first partitioned into eight cohorts based on the husband's birth~~
25 ~~year (prior to 1850, 1850-1875, 1876-1900, and 1901-1925) and sex. For the time~~
26 ~~since bereavement analyses, we did not partition into cohorts because the number~~
27 ~~of widow(er)s who died within 6 months after being bereaved is small (N=224).~~
28 ~~Characteristics of these couples by husband birth cohort are shown in Table 1.~~

Statistical analyses

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45 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
46 Hazards (CPH) model where the response variable or survival time is 'age at
47 death' and the event is 'death'. The model is used to study the association of
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widowhood and mortality rates in the surviving spouse, while adjusting for covariates such as education, health habits, age in years, number of children, and remarriage. In some of our analyses, we adjusted for remarriage and number of children as covariates; we did not adjust for education or health habits. Husbands and wives were always analyzed separately. Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional where the response variable or survival time 'age at widowhood or death' and the event is 'death'. The model is used to study the association of widowhood and mortality rates in the surviving spouse, while adjusting for covariates such as education, health habits, age in years, number of children, and remarriage. In some of our analyses, we adjusted for remarriage and number of children as covariates; we did not adjust for education or health habits. Widows and widowers were always analyzed separately.

The main covariate, widowhood, was monitored as a time-dependent covariate, by assigning a value of 1 for each time period the individual was widowed and 0 otherwise. The remarriage covariate was also a time-dependent covariate, by assigning a value 1 for the widowed period if the individual remarried and 0 otherwise. The number of surviving children was a time-independent covariate and counted at the beginning of widowhood period of the surviving spouse. The covariate was divided into three categories, as follows: 0=number of surviving children is ≤ 2 , 1=number of surviving children is between 3 and 6, and 2=number of surviving children is >6 . The categories ≤ 2 children, 3-

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9 6 children, and > 6 children are separate. These boundaries were chosen to ensure
10 categories that were roughly balanced in size. To evaluate any possible bias by
11 counting the number of surviving children at the death of the first spouse, we also
12 estimated how many couples changed categories between the death of the first
13 spouse and the death of second spouse due to death of one or more children
14 during the widowhood period. The number of surviving children was a time-
15 independent covariate and divided into categories, as follows: 0=number of
16 surviving children is ≤ 2 , 1=number of surviving children is between 3 and 6, and
17 2=number of surviving children is > 6 . The categories ≤ 2 children, 3-6 children, and
18 > 6 children are separate. These boundaries were chosen to ensure categories that were
19 roughly balanced in size.

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30 Survival analyses were performed using the R programming environment.¹⁷
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32 All estimates and confidence intervals were obtained using the `coxph` function
33 available in the `survival` package.

34 35 36 **Representation of survival data for CPH analysis**

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38 We show below examples of how we represented the survival data for CPH
39 analysis using a time modeling approach. To represent the survival data columns, the
40 standard approach is to convert the couple's demographic information, date of
41 death, date of birth, remarriage and number of surviving children into columns
42 representing **start** time, **stop** time, **event** status, and all of the included covariates
43 representing **start** time, **stop** time, **event** status, and all of the included covariates
44 (widowhood, remarriage, and number of surviving children).^{18 19} Here (**start**,
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47 **stop**] is an interval of risk, open on the left and closed on the right; the **event**
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9 column is set to 1 if the subject had an event (death) at time **stop** and is 0
10 otherwise. To illustrate the subtleties, we consider a hypothetical example of
11 three wives (ID1, ID2 and ID3) from the cohort 1850-1875.
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16 • *ID*₁: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
17 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
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22 • *ID*₂: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
23 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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28 • *ID*₃: born on 03/01/1871; never widowed and never remarried; number of
29 surviving children = 4 and died on 04/01/1930 at age 59.
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34 The data used for the CPH considering widowhood as a single time-
35 dependent covariate is presented in Table 2a. The following model was used to
36 estimate the association between widowhood and mortality.
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$$h(t) = h_0(t) \exp[\beta W]$$

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41 This model can be run with additional covariates and results that adjust for the
42 ability to remarry and the number of surviving children are presented.
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45 **Timing of widowhood**

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47 The above model considers widowhood as a single time-dependent
48 covariate, evaluating the association between mortality and being
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9 widowed. One can further ask if that association varies over the life
10 course by considering the impact of age at widowhood on mortality.
11 We addressed this issue following the approach of Mineau et al. by
12 adding terms for age at widowhood into the model as covariates (<45,
13 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to
14 age at widowhood (see Table 2b).⁸ These terms were included in the model
15 as time-dependent dummy variables, created to represent the
16 widowhood experience of each individual across the 5 age windows
17 spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} .

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26 Further, we also addressed the impact of remarriage and number of
27 surviving children on the mortality of Amish individuals.~~Further, we~~
28 ~~also addressed the impact of remarriage and number of surviving~~
29 ~~children on mortality of widowed individuals.~~
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34 The following model can be used to estimate the association
35 between each of the five dummy variables and mortality:

$$36 \quad h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

37
38 where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} ,
39 W_{65-74} , W_{75+} columns provided in the Table 2b.

40 41 42 43 **Time since bereavement**

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45 Next, we evaluated the association between mortality and time since bereavement
46 or widowhood. We followed the approach of Schaefer et al. by considering the
47 following time since bereavement ranges: 0-6 months, 7-12 months, 13-24
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9 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This
10 approach allowed us to estimate HR according to time since bereavement (see
11 Table 2c). The columns TSB₀₋₆, TSB₇₋₁₂, ... in Table 2c are time dependent
12 covariates that change with the survival time associated with widowed husbands
13 and wives. We did not account for remarriage in this analysis because of missing
14 remarriage dates.
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20 The following model was used to study the association between mortality and
21 time since bereavement:
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$$23 \quad h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

24 where W_1, \dots, W_7 dummy variables are associated with the TSB₀₋₆, TSB₇₋₁₂,
25 TSB₁₃₋₂₄, TSB₂₅₋₃₆, TSB₃₇₋₄₈, TSB₄₉₋₆₀ and TSB_{>60} columns provided in
26 the Table 2c.
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33 RESULTS

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37 We initially partitioned the 10,892 couples from AGDB into three cohorts ranging
38 in size from 3,210 (husband birth year 1850-1875) to 3,971 (husband birth year
39 1876-1900). Wives were more likely to die after their husbands in all three
40 cohorts while husbands had a higher mean age at death than their wives. The
41 average age differences between the husbands and wives for all cohorts are shown
42 in Table 1. The number of remarried wives was far smaller than the number of
43 remarried husbands (n = 1,290 widowed husbands vs 378 widowed wives). The
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9 number and proportion of remarried husbands and wives were increased in the
10 more contemporary cohorts (see Table 1). In contrast to other populations from
11 the 19th and early 20th centuries,⁴ the majority of widowed husbands did not
12 remarry, making it interesting to study the effect of remarriage in the Amish
13 population.~~We initially partitioned the 15,611 couples from AGDB into four~~
14 ~~cohorts ranging in size from 3,210 (husband birth year 1850-1875) to 4,466~~
15 ~~(husband birth year 1876-1900). Wives were more likely to die after their~~
16 ~~husbands in all cohorts, although in the three earliest cohorts (prior to 1850,~~
17 ~~1850-1875, and 1876-1900), husbands had a higher mean age at death than their~~
18 ~~wives. The average age differences between the husbands and wives for all~~
19 ~~cohorts are shown in Table 1. The number of remarried wives was far smaller~~
20 ~~than the number of remarried husbands (n = 1,943 widowed husbands vs 613~~
21 ~~widowed wives). The number and proportion of remarried husbands and wives~~
22 ~~were increased in the more contemporary cohorts (see Table 1). In contrast to~~
23 ~~other populations from the 19th and early 20th centuries,⁴ the majority of widowed~~
24 ~~husbands did not remarry, making it interesting to study the effect of remarriage~~
25 ~~in the Amish population.~~

41 **Hazard Ratios**

42 ~~The overall hazard ratios (HR), and their 95% confidence intervals (CIs),~~
43 ~~associated with widowhood are displayed in Figure 2 according to birth cohort.~~
44 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
45 associated with widowhood are displayed in Figure 2 according to birth cohort.

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9 The significance of these hazard ratios are indicated with the help of p-values
10 (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2.

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12 These hazard ratios were estimated using the CPH model and design provided in
13 Table 2a. As expected, widowhood was associated with increased mortality for
14 both husbands and wives following the death of spouse, with HRs ranging
15 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
16 husbands in cohort 1850-1875). The impact of widowhood was
17 disproportionately greater on surviving husbands than on surviving wives in the
18 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
19 between widowed husbands vs. wives in the 1850-1875. The impact of
20 widowhood was disproportionately greater on surviving husbands than on
21 surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little
22 difference in mortality between widowed husbands vs wives in the 1850-1875 and
23 1901-1925 cohorts. There was no clear trend showing that the hazard ratios
24 changed across cohorts in either surviving husbands or wives. Since no clear
25 differences were observed between birth cohorts, all birth cohorts were combined
26 to estimate the association between age at widowhood and mortality.

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41 Figure 3 shows the hazard ratios (and 95% CIs) for the model in which age at
42 widowhood is accounted for (design model in Table 2b). Figure 3 shows the
43 hazard ratios along with 95% CIs, and range of p-values for the model in which
44 age at widowhood is accounted for (design model in Table 2b). These results
45 reveal markedly higher bereavement effects in each age at widowhood group
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9 compared to the results shown in Figure 2, consistent with the notion that the
10 bereavement effect may be diminished over time and most pronounced in the
11 years proximal to the death of the spouse.^{2 5 20 21} In nearly all age at widowhood
12 categories, the bereavement effect is stronger in widowed husbands than in
13 widowed wives.
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19 The number of children surviving when the first spouse died (with two
20 pairwise comparisons 3-6 vs ≤ 2 ; > 6 vs ≤ 2) was included as a time-independent
21 covariate in the models whose results are shown in Figure 3. Number of surviving
22 children (> 6 vs ≤ 2) was included as a covariate in the models whose results are
23 shown in Figure 3. In general, there was a very weak association between number
24 of surviving children and mortality in husbands and wives. Contrary to our
25 expectations and a prior study,⁸ in each case, a higher number of surviving
26 children was not significantly associated with lower mortality in both husbands
27 and wives (See Figure 3). Further, the results in Figure 3 show that the effect of
28 bereavement decreases if the Amish individual remarries.
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38 There are two potential sources of bias in analyses involving the number of
39 surviving children covariate. Couples in which one spouse died at age < 50 are
40 likely not to have as many children as couples in which both parents survived to
41 at least age 50. To quantify this bias, we repeated the analysis in Figure 3 by
42 excluding all the couples who got widowed before age 50 (data not shown). The
43 results show that there is no significant change in the hazard ratios vs. Figure 3.
44 The maximum change for any of the hazard ratios related to the number of
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9 surviving children was 0.03 (data not shown).

10 On the other end of the age spectrum, there is a potential source of bias as
11 more children may die, the longer the surviving parent lives. Because the number
12 of children was divided into three categories ≤ 2 , 3-6, and > 6 , any possible bias
13 could arise when and only when the death of a child shifted the number of
14 surviving children from a higher category to a lower category. Over all couples,
15 the number of widows or widowers whose number of surviving children changed
16 to a lower category from the date of widowhood to the date of death was only 298
17 (< 3% of all couples).

18 When the analyses corresponding to Figure 3 were repeated excluding these
19 298 couples, the hazard ratios were essentially unchanged. The maximum change
20 was 0.04; sometimes the hazard ratio increased slightly and sometime it decreased
21 slightly (data not shown). In general, there was a very weak association between
22 number of surviving children and mortality in widowed husbands and widowed
23 wives. Contrary to our expectations and a prior study,⁸ in each case, a higher
24 number of surviving children was associated with higher mortality in the
25 widowed husband/wife. When analyses were repeated without the number of
26 surviving children as a covariate, the estimates of the hazard ratios were
27 essentially unchanged (data not shown). Further, the results in Figure 3 show that
28 the effect of bereavement decreases if the widowed individual remarries.

29 Hazard ratios and 95% CIs for the time since bereavement analysis are shown
30 in Figure 4. The significant p-values are indicated in Figure 4. These results were

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obtained using the CPH model and the design defined in Table 2c. The results show that there is an increase in risk of mortality for recently widowed husbands and wives, and the hazard decreases with time since bereavement but remains significantly greater than 1. Further, the hazard is higher (not significant) in wives vs. husbands during the first 12 months following bereavement. Hazard ratios and 95% CIs for the time since bereavement analysis are shown in Figure 4. These results were obtained using the CPH model and the design defined in Table 2c. The results show that there is an increase in risk of mortality for recently widowed husbands and wives, and the hazard decreases with time since bereavement but remains significantly greater than 1. Further, the hazard is higher (not significant) in wives vs husbands during the first 12 months following bereavement. To address the issue that many individuals in the 1901-1925 cohort may still have been alive at the last AGDB update and hence censored, we performed the analysis for time since bereavement omitting the cohort 1901-1925. The results show that the hazard ratios provided in Figure 4 have not changed significantly; for example, for 25-26 months post bereavement the hazard ratios change from 1.30 and 1.23 for all cohort to 1.25 and 1.17 for the three eldest cohorts.

The range of p-values provided in Figures 2-4 indicates the significance of each of the hazard ratios. For example, in Figure 4, the p-value = 0.0001 associated with the hazard ratio = 1.39 (males; time since bereavement range < 6 months) strongly indicates that the hazard ratio is significantly > 1. Similarly, the p-value = 0.039 associated with the hazard ratio = 1.15 (males; time since

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9 bereavement 37-48 months) weakly indicates the significance of the hazard ratio
10 > 1.

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12 Graphical checks of the overall adequacy of the CPH models were
13 performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a
14 reasonable fit to the data (data not shown). The *deviance residual* plots
15 revealed no obvious outliers in the data (data not shown). Further, the Wald test
16 statistic was used to test the fit of the final model,¹⁸ and according to this test
17 statistic, the final model fits the data reasonably well (Supplementary Tables S1,
18 S2, and S3).
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27 **DISCUSSION**

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31 This is the first comprehensive study to evaluate the relationship between
32 bereavement and social support in the Amish population. This study provides
33 evidence that Amish widows and widowers have increased mortality risk
34 compared to married cohort members. Although it is difficult to determine
35 whether this effect is of equivalent magnitude as that observed in studies of other
36 populations, the most recent studies of the bereavement effect using the CPH
37 suggest that our findings are generally consistent with data from other
38 populations.⁴
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46 Several previous studies on American, European, and Middle Eastern
47 populations have found that mortality is magnified in individuals widowed at a
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9 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}

10 The LDS study is closest to our study because of the large family sizes and the
11 population selected as a religious isolate.⁸ The effects of bereavement on
12 mortality with respect to gender and the age at widowhood ranges observed in the
13 Amish are also largely consistent with those observed in the LDS population,⁸ and
14 other populations such as Finland and Israel (Figure 3).^{6 22} As expected, widowers
15 showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure
16 2).
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18 In the present study, the association between bereavement and mortality is
19 greater in the first 6 months for both men and women (Figure 4), consistent with
20 previous findings.^{2 3 5 20 21 23} The mortality risks in the first 6 months are lower in
21 the Amish (Figure 4) compared to some studies,^{2 5 20 21} but not all studies.^{3 23} One
22 common pattern observed in this and other studies is that the initially high
23 bereavement effect first decreases but then increases with time since
24 bereavement.^{3 23} We speculate that the increased mortality during the first 6
25 months might reflect acute effects related to the loss of a spouse, while the
26 gradual increases in mortality emerging in later life might reflect decreased
27 survival from aging-related diseases that is unmasked in the absence of spousal
28 support.
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30 One strength of our study is that we could evaluate the effect of remarriage
31 because there were substantial numbers of widowed individuals who did and did
32 not get remarried (Table 1) Two strengths of our study are that, unlike many of the
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9 previously published studies,^{5 13 16 20 21 24} we did not incorporate censoring
10 methods into our analysis because of the high availability of death dates of the
11 Amish widows and widowers in the first three cohorts; and we could study the
12 effect of remarriage because there were substantial numbers of widowed
13 individuals who did and did not get remarried (Table 1). Shor et al. noted that the
14 difficulty in studying the effect of remarriage in populations where all individuals
15 are deceased because “In previous decades, widowed men almost always
16 remarried”.⁴ As suggested by previous studies,^{8 9} remarriage after the death of a
17 spouse significantly influences the bereavement effect because it is associated
18 with increased survival of both males and females across all cohorts . This
19 association is likely influenced by the fact that males or females who get
20 remarried are in sufficiently good physical and mental health to do so. Further,
21 the survival rate is higher for males as compared to females, possibly reflecting
22 support in Amish society for males getting remarried.As suggested by previous
23 studies;⁸⁻⁹ remarriage after the death of a spouse significantly influences the
24 bereavement effect because it is associated with increased survival of both
25 widowers and widows across all cohorts and age at widowhood ranges. This
26 association is likely influenced by the fact that widowers or widows who get
27 remarried are in sufficiently good physical and mental health to do so. Further,
28 the survival rate is higher for widowers as compared to widows, possibly
29 reflecting support in Amish society for males getting remarried.

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48 Interestingly, increasing numbers of surviving children at the time of
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9 widowhood did not confer a survival advantage for Amish individuals. This result
10 was counter to our hypothesis that children can help provide social support for
11 their parents. The hazard ratio was greater than 1.0 (but not significant) for all
12 Amish individuals with number of surviving children > 6 as compared to ≤
13 2. Interestingly, increasing numbers of surviving children at the time of
14 widowhood did not confer a survival advantage for Amish widows or widowers.
15 This result was counter to our hypothesis that children can help provide social
16 support for their parents. The hazard ratio was greater than 1.0 (but not
17 significant) for all widowers and widows with number of surviving children > 6
18 as compared to ≤ 2. Spouses in the Amish society may also provide unique
19 emotional, psychological, and social support to each other which cannot be
20 provided by their surviving children. The lack of protective association was
21 similarly observed when the number of surviving children was considered as a
22 linear or as a categorical variable (data not shown). This contrasts with data from
23 the Utah Population Database, in which increasing numbers of children were
24 associated with a decreased hazard ratio.⁸

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41 The implication of these findings is that differential familial support
42 following bereavement does not appear to be the key factor affecting mortality
43 increases in widowed Amish populations. One potential explanation is that
44 spouses, as “attachment figures,” provide a unique, irreplaceable social support
45 which must be considered independently from other ancillary support providers,
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9 such as children.²⁵⁴ Data suggesting that relative mortality risk is also elevated in
10 divorced and never-married populations may support this hypothesis.³ In addition,
11 quality of social support has significantly greater effects on well-being than
12 quantity of support in elderly populations of both sexes.²⁶⁵

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16 In adults aged 65 and older, lower reported social support has been associated
17 with decreased life satisfaction and increased depressive symptoms.²⁷⁶ Family
18 members and close friends should be especially vigilant during the sensitive acute
19 period following the loss of a spouse, when relative mortality risk is the highest.
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24 ~~A limitation of our data is that causes of death were unavailable. Future~~
25 ~~research is needed to determine whether differential measures of social support~~
26 ~~may be associated with certain causes of death in widowed populations. Divorce~~
27 ~~in the Amish is sufficiently rare that this was not a major potential source of error.~~
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29 There are two limitations of our data. First, the number of surviving children
30 covariate was simplified because it was treated as a time-independent covariate,
31 although some children die before their parents, so it could be considered as time-
32 dependent instead. Second, the causes of death were unavailable. Future research
33 is needed to study the effect of the loss of a child on the mortality of husbands and
34 wives by considering the number of surviving children as a time-dependent
35 covariate. Also, research is needed to determine whether differential measures of
36 social support may be associated with certain causes of death in widowed
37 populations. Divorce in the Amish is sufficiently rare that this was not a major
38 potential source of error.
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9 Our study results indicate that remarriage plays an important role in
10 improving the survival rate of males and females in the Amish population.
11 Contrary to results from previous studies, an increase in the number of surviving
12 children was associated with a decreased survival rate of Amish individuals.~~Our~~
13 ~~study results indicate that remarriage plays an important role in improving the~~
14 ~~survival rate of widows and widowers in the Amish population. Contrary to~~
15 ~~results from previous studies, an increase in the number of surviving children was~~
16 ~~associated with a decreased survival rate of widowed individuals.~~

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25 to improve the manuscript.

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27
28 **Contributors** BDM conceived the study. AS and SS did the research and data
29 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
30 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
31 managed the project with assistance from BDM. AAS manages the AGDB project
32 at the National Institutes of Health and KAR managed the AGDB data at
33 University of Maryland Medical School during the period of this project. ARS
34 provided resources and advice. All authors proofread and approved the
35 manuscript.

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9 Geriatric Research and Education Center, Baltimore Veterans Administration
10 Medical Center.

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13 **Competing interests** None.

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16 **Ethics approval** The construction and maintenance of AGDB are covered under
17 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
18 Investigator.
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22 **Provenance and peer review** Not commissioned; externally peer reviewed.
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25 **Data sharing statement** Data from the AGDB are available to investigators
26 (including ARS and various others not participating in this study) who have an
27 IRB-approved protocol to study the Amish or other Anabaptist groups.
28 Investigators can request access to AGDB by writing to AAS and to Dr.
29 Biesecker.
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FIGURE LEGENDS

Figure 1. A flow diagram that represents all the steps performed for filtering 10,892 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. "Unknown spouse" refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. "Birth year too late" means that the birth year of the husband or wife is known and is > 1901. "Dates not recognized by R" are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. "Implausible birth or death dates" refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

~~Figure 1. A flow diagram which represents all the steps performed for filtering 15,611 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. "Unknown spouse" refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. "Birth year too late" means that~~

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9 the birth year of the husband is known and is > 1925 . “Dates not recognized by
10 R” are invalid dates such as the 31st of June, which got into AGDB due to errors
11 in the original sources. “Implausible birth or death dates” refers to a few
12 individuals who are shown as married but have lifespans of less than 10 years
13 likely due to typos in the birth year in the original sources.
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19 Figure 2. Hazard ratios of widowed husbands and wives versus their married
20 counterparts (design provided in Table 2a); * and ** on the top of the blocks
21 represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001 ,
22 respectively. ~~Figure 2. Hazard ratios of widowed husbands and wives versus their~~
23 ~~married counterparts (design provided in Table 2a).~~
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28 Figure 3. Hazard ratios of widowed husbands and wives versus their married
29 counterparts according to age at widowhood (design provided in Table 2b);
30 NSC1: Number of Surviving Children (3-6 vs ≤ 2); NSC2: Number of Surviving
31 Children (>6 vs ≤ 2); * and ** on the top of the blocks represent significance of
32 hazard ratios with p-value < 0.05 and p-value < 0.001 , respectively. ~~Figure 3.~~
33 ~~Hazard ratios of widowed husbands and wives versus their married counterparts~~
34 ~~according to age at widowhood. (design provided in Table 2b).~~
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42 Figure 4. Hazard ratios of widowed husbands and wives versus their married
43 counterparts according to time since bereavement (months) (design provided in
44 Table 2c); * and ** on the top of the blocks represent significance of hazard ratios
45 with p-value < 0.05 and p-value < 0.001 , respectively. ~~Figure 4. Hazard ratios of~~
46 ~~widowed husbands and wives versus their married counterparts according to time~~
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since bereavement (months). (design provided in Table 2c).

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TABLES

Table 1: Characteristics of 10,892 spouse pairs according to birth cohort of husband
 1: Characteristics of 15,611 spouse pairs according to birth cohort of husband

	Cohorts			
	Pre-1850	1850-1875	1876-1900	1901-1925
Number of couples	3711	3210	4466	4224
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2360(53.0)	2439(57.7)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	2106(47.0)	1785(42.3)
Mean husband age at widowhood	63.0	59.8	62.2	67.9
Mean wife age at widowhood	62.0	62.8	66.5	69.6
Mean widowed husband survival in years	14.4	18.3	18.6	15.0
Mean widowed wife survival in years	15.3	16.0	17.0	15.1
Mean husband age at death	71.1	72.0	74.6	76.2
Mean wife age at death	69.6	68.5	72.7	77.0
Mean age difference husband-wife	3.4	2.8	2.1	1.3
Mean number of children	5.4	5.3	5.3	5.8
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	13(33.8)	588(33.0)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	221(9.4)	194(8.0)

Cohorts	Pre-1850	1850-1875	1876-1900
Number of couples	3711	3210	3971
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2043(51.4)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	1928(48.2)
Mean husband age at widowhood	63.0	59.8	62.2
Mean wife age at widowhood	62.0	62.8	66.9
Mean widowed husband survival in years	14.4	18.3	18.4
Mean widowed wife survival in years	15.3	16.0	16.6
Mean husband age at death	71.1	72.0	74.6
Mean wife age at death	69.6	68.5	72.6

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Mean age difference husband-wife	3.4	2.8	2.0
Mean number of children	5.4	5.3	5.3
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	648(33.6)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	180(8.8)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0
ID ₁	51	52	0	0	0	0	0	0	1	0
ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

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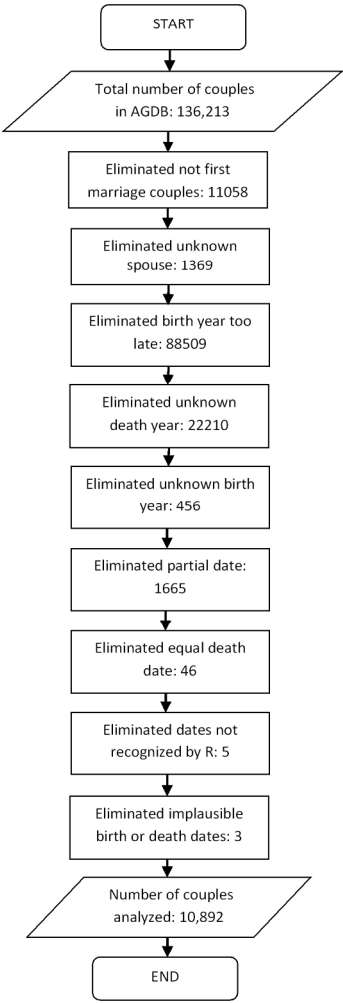
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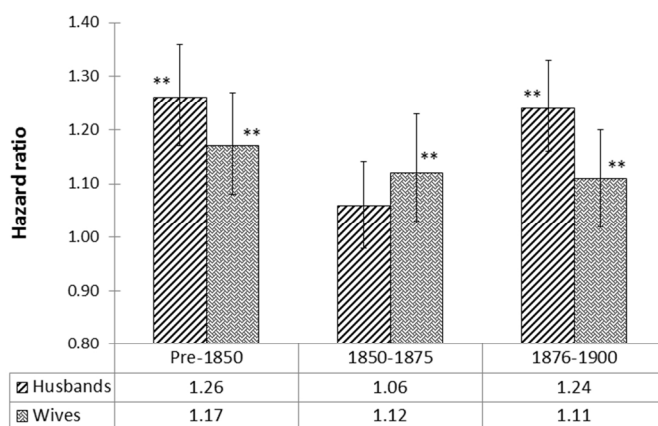
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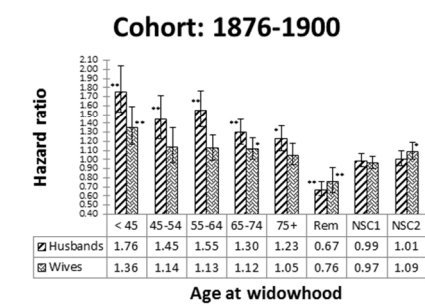
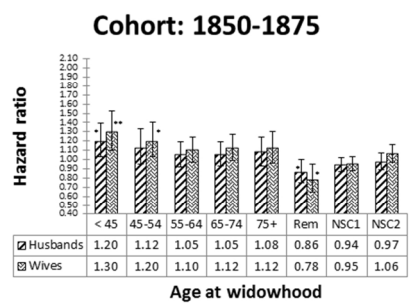
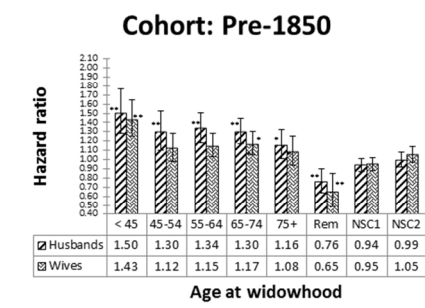
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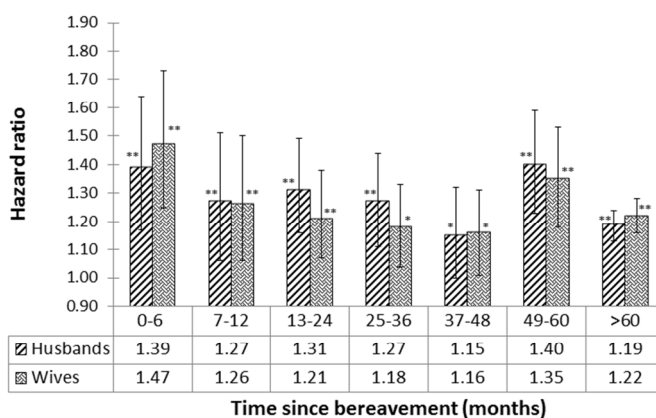


Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months) (design provided in Table 2c); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.
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Table S1: Wald statistics associated with models that are used to study the association between widowhood and mortality.

Cohorts	Pre-1850	1850-1875	1876-1900
Husbands	38.2 ^{**}	5.9 [*]	38.0 ^{**}
Wives	15.2 ^{**}	6.9 [*]	6.8 [*]
* P<0.05 and ** P<0.001			

Table S2: Wald statistics associated with models that are used to study the association between timing of widowhood and mortality.

Cohorts	Pre-1850	1850-1875	1876-1900
Husbands	57.2 ^{**}	15.8 [*]	87.7 ^{**}
Wives	39.8 ^{**}	20.3 ^{**}	28.9 ^{**}
* P<0.05 and ** P<0.001			

Table S3: Wald statistics associated with models that are used to study the association between time since bereavement and mortality.

Husbands	94.6 ^{**}
Wives	84.3 ^{**}
** P<0.001	



Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Manuscript ID:	bmjopen-2013-003670.R3
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Keywords:	EPIDEMIOLOGY, GERIATRIC MEDICINE, MENTAL HEALTH, PUBLIC HEALTH

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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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Author-suggested Keywords: bereavement effect, Amish, remarriage, social support, Cox proportional hazards, time-dependent covariate

Word Counts:

Abstract: 248

Introduction through Discussion: 3,757.

ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 10,892 Amish couples born during 1725-1900 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure The survival time is 'age'; event is 'death'. Hazard ratios of widowed individuals with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both males and females. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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3 socially cohesive Amish community. Remarriage is associated with a significant
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5 decrease in the mortality risk among Amish individuals. Contrary to results from
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7 previous studies, an increase in the number of surviving children was associated
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9 with decreased survival rate.
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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and the mortality of a surviving spouse in the Amish population.
- We evaluated the association of bereavement and mortality of a surviving spouse with respect to gender, age at widowhood, and time since bereavement while accounting for remarriage, and number of surviving children using Cox proportional hazard models.

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks among Amish individuals, and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.
- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.

- The number of surviving children was simplified to be a time independent covariate although it may change between the death of the first parent and the death of the second parent.

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INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.^{2 5} Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6 7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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The Amish maintain a cultural identity distinct from mainstream American

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3 culture that is characterized by their traditional dress, a plain lifestyle, and non-
4 adoption of modern technology (e.g., electricity, cars, telephones), German
5 dialect, separate school system, and ultra-conservative Anabaptist religious
6 practices. A central tenet of Amish culture throughout their history in the USA
7 has been social cohesiveness with emphasis on family and community. Members
8 of this tight-knit society have extraordinary social support from cradle to grave,
9 including community-financing of medical costs and support during times of
10 need. The community financing of medical costs became increasingly formalized
11 during the 20th century to the point where it is now a formal system of self-
12 insurance called “Amish Aid”.
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27 To gain a different perspective on potential influences of social support on
28 bereavement, we assessed the bereavement effect in an Amish population, whose
29 culture is characterized by its core beliefs in community and social cohesion.
30 Examples of the strong degree of social support within the Amish include
31 community-managed health insurance and community ties through membership in
32 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
33 We reasoned that the high level of social support in the Amish population might
34 mitigate the bereavement effect. Previous studies used covariates such as
35 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
36 neighborhood interaction, and household size to study the relationship between
37 bereavement and social support.^{4 8 13}
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53 We estimated bereavement effects in widows and widowers separately, and
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3 further assessed mortality risks as a function of age at widowhood and time since
4 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
5 association of bereavement and mortality of widowed husbands and wives. We
6 considered remarriage and number of surviving children as additional potential
7 modifiers of the bereavement effect.
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14 15 16 17 **MATERIAL AND METHODS**

18 19 **Data source**

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21 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
22 effect of bereavement on the Amish population.^{14 15} The AGDB is a
23 computerized genealogy database of Amish and some Mennonite populations, last
24 updated in 2010. This database includes information about the family
25 relationships and the birth, marriage, and death dates of children of Amish
26 individuals from North America, mostly located in Pennsylvania, Ohio, and
27 Indiana. The "individual table" of AGDB contains information about 539,822
28 individuals. The "relationship table" includes information about 136,213 Amish
29 couples. An individual who is married multiple times participates in multiple
30 relationship table entries. There are 1,369 relationship entries among the 136,213
31 entries concerning children for whom one or both biological parents are unknown
32 (Figure 1).
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50 Amish people rarely give birth out of wedlock, and a vast majority of the
51 mating samples we used are documented as marriages in the three original sources
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3 from which AGDB is derived. However, there is less availability of marriage
4 dates than birth dates; we did not wish to exclude couples with missing marriage
5 dates. For this study, we included couples in which both spouses met four
6 conditions: (1) they were born prior to January 1, 1901; (2) their birth and death
7 dates were recorded; (3) they were in their first marriage; and (4) they did not die
8 on the same day. Constraint (4) excludes couples who may have died from a
9 shared disaster (e.g., accident). A total of 10,892 couples met the inclusion
10 criteria. The oldest person included was born on February 14, 1725, and the
11 youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were
12 applied.
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27 Our data set included 10,892 couples with information on their date of birth,
28 date of death, number of surviving children, family IDs, and remarriage status
29 after widowhood. For some analyses, these couples were first partitioned into
30 three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and
31 based on both husband's and wife's birth year (1876-1900) and sex. For the time
32 since bereavement analyses, we did not partition into cohorts because the number
33 of widow(er)s who died within 6 months after being bereaved is small (N=291).
34 Characteristics of these couples are shown in Table 1.
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47 **Statistical analyses**

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49 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
50 Hazards (CPH) model where the response variable or survival time is 'age at
51 death' and the event is 'death'. The model is used to study the association of
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3 widowhood and mortality rates in the surviving spouse, while adjusting for
4 covariates such as education, health habits, age in years, number of children,
5 and remarriage. In some of our analyses, we adjusted for remarriage and
6 number of children as covariates; we did not adjust for education or health
7 habits. Husbands and wives were always analyzed separately.

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15 The main covariate, widowhood, was monitored as a time-dependent
16 covariate, by assigning a value of 1 for each time period the individual was
17 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
18 covariate, by assigning a value 1 for the widowed period if the individual
19 remarried and 0 otherwise. The number of surviving children was a time-
20 independent covariate and counted at the beginning of widowhood period of the
21 surviving spouse. The covariate was divided into three categories, as follows:
22 0=number of surviving children is ≤ 2 , 1=number of surviving children is between
23 3 and 6, and 2=number of surviving children is > 6 . The categories ≤ 2 children, 3-
24 6 children, and > 6 children are separate. These boundaries were chosen to ensure
25 categories that were roughly balanced in size. To evaluate any possible bias by
26 counting the number of surviving children at the death of the first spouse, we also
27 estimated how many couples changed categories between the death of the first
28 spouse and the death of second spouse due to death of one or more children
29 during the widowhood period.

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51 Survival analyses were performed using the R programming environment.¹⁷
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53 All estimates and confidence intervals were obtained using the `coxph` function
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6 **Representation of survival data for CPH analysis**

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8 We show below examples of how we represented the survival data for CPH
9 analysis using a time modeling approach. To represent the survival data columns, the
10 standard approach is to convert the couple's demographic information, date of
11 death, date of birth, remarriage and number of surviving children into columns
12 representing **start** time, **stop** time, **event** status, and all of the included covariates
13 (widowhood, remarriage, and number of surviving children).^{18 19} Here (**start**,
14 **stop**] is an interval of risk, open on the left and closed on the right; the **event**
15 column is set to 1 if the subject had an event (death) at time **stop** and is 0
16 otherwise. To illustrate the subtleties, we consider a hypothetical example of
17 three wives (ID1, ID2 and ID3) from the cohort 1850-1875.
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- 34 • *ID*1: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
35 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
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- 40 • *ID*2: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
41 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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- 48 • *ID*3: born on 03/01/1871; never widowed and never remarried; number of
49 surviving children = 4 and died on 04/01/1930 at age 59.
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3 The data used for the CPH considering widowhood as a single time-
4 dependent covariate is presented in Table 2a. The following model was used to
5 estimate the association between widowhood and mortality.
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$$10 \quad h(t) = h_0(t) \exp[\beta W]$$

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12 This model can be run with additional covariates and results that adjust for the
13 ability to remarry and the number of surviving children are presented.
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17 **Timing of widowhood**

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19 The above model considers widowhood as a single time-dependent
20 covariate, evaluating the association between mortality and being
21 widowed. One can further ask if that association varies over the life
22 course by considering the impact of age at widowhood on mortality.
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24 We addressed this issue following the approach of Mineau et al. by
25 adding terms for age at widowhood into the model as covariates (<45,
26 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to
27 age at widowhood (see Table 2b).⁸ These terms were included in the model
28 as time-dependent dummy variables, created to represent the
29 widowhood experience of each individual across the 5 age windows
30 spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} .
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32 Further, we also addressed the impact of remarriage and number of
33 surviving children on the mortality of Amish individuals.
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51 The following model can be used to estimate the association
52 between each of the five dummy variables and mortality:
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$$h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , W_{75+} columns provided in the Table 2b.

Time since bereavement

Next, we evaluated the association between mortality and time since bereavement or widowhood. We followed the approach of Schaefer et al. by considering the following time since bereavement ranges: 0-6 months, 7-12 months, 13-24 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This approach allowed us to estimate HR according to time since bereavement (see Table 2c). The columns TSB_{0-6} , TSB_{7-12} , ... in Table 2c are time dependent covariates that change with the survival time associated with widowed husbands and wives. We did not account for remarriage in this analysis because of missing remarriage dates.

The following model was used to study the association between mortality and time since bereavement:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

where W_1, \dots, W_7 dummy variables are associated with the TSB_{0-6} , TSB_{7-12} , TSB_{13-24} , TSB_{25-36} , TSB_{37-48} , TSB_{49-60} and $TSB_{>60}$ columns provided in the Table 2c.

RESULTS

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3 We initially partitioned the 10,892 couples from AGDB into three cohorts ranging
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5 in size from 3,210 (husband birth year 1850-1875) to 3,971 (husband birth year
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7 1876-1900). Wives were more likely to die after their husbands in all three
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9 cohorts while husbands had a higher mean age at death than their wives. The
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11 average age differences between the husbands and wives for all cohorts are shown
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13 in Table 1. The number of remarried wives was far smaller than the number of
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15 remarried husbands (n = 1,290 widowed husbands vs 378 widowed wives). The
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17 number and proportion of remarried husbands and wives were increased in the
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19 more contemporary cohorts (see Table 1). In contrast to other populations from
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21 the 19th and early 20th centuries,⁴ the majority of widowed husbands did not
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23 remarry, making it interesting to study the effect of remarriage in the Amish
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25 population.
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31 32 **Hazard Ratios**

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34 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
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36 associated with widowhood are displayed in Figure 2 according to birth cohort.
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38 The significance of these hazard ratios are indicated with the help of p-values
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40 (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2.
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42 These hazard ratios were estimated using the CPH model and design provided in
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44 Table 2a. As expected, widowhood was associated with increased mortality for
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46 both husbands and wives following the death of spouse, with HRs ranging
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48 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
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50 husbands in cohort 1850-1875). The impact of widowhood was
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3 disproportionately greater on surviving husbands than on surviving wives in the
4 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
5 between widowed husbands vs. wives in the 1850-1875. There was no clear trend
6 showing that the hazard ratios changed across cohorts in either surviving
7 husbands or wives.
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Figure 3 shows the hazard ratios along with 95% CIs, and range of p-values for the model in which age at widowhood is accounted for (design model in Table 2b). These results reveal markedly higher bereavement effects in each age at widowhood group compared to the results shown in Figure 2, consistent with the notion that the bereavement effect may be diminished over time and most pronounced in the years proximal to the death of the spouse.^{2 5 20 21} In nearly all age at widowhood categories, the bereavement effect is stronger in widowed husbands than in widowed wives.

The number of children surviving when the first spouse died (with two pairwise comparisons 3-6 vs ≤ 2 ; > 6 vs ≤ 2) was included as a time-independent covariate in the models whose results are shown in Figure 3. In general, there was a very weak association between number of surviving children and mortality in husbands and wives. Contrary to our expectations and a prior study,⁸ in each case, a higher number of surviving children was not significantly associated with lower mortality in both husbands and wives (See Figure 3). Further, the results in Figure 3 show that the effect of bereavement decreases if the Amish individual remarries.

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There are two potential sources of bias in analyses involving the number of surviving children covariate. Couples in which one spouse died at age <50 are likely not to have as many children as couples in which both parents survived to at least age 50. To quantify this bias, we repeated the analysis in Figure 3 by excluding all the couples who got widowed before age 50 (data not shown). The results show that there is no significant change in the hazard ratios vs. Figure 3. The maximum change for any of the hazard ratios related to the number of surviving children was 0.03 (data not shown).

On the other end of the age spectrum, there is a potential source of bias as more children may die, the longer the surviving parent lives. Because the number of children was divided into three categories ≤ 2 , 3-6, and > 6 , any possible bias could arise when and only when the death of a child shifted the number of surviving children from a higher category to a lower category. Over all couples, the number of widows or widowers whose number of surviving children changed to a lower category from the date of widowhood to the date of death was only 298 (< 3% of all couples).

When the analyses corresponding to Figure 3 were repeated excluding these 298 couples, the hazard ratios were essentially unchanged. The maximum change was 0.04; sometimes the hazard ratio increased slightly and sometime it decreased slightly (data not shown).

Hazard ratios and 95% CIs for the time since bereavement analysis are shown in Figure 4. The significant p-values are indicated in Figure 4. These results were

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obtained using the CPH model and the design defined in Table 2c. The results show that there is a high risk of mortality for recently widowed husbands and wives. Further, the hazard is higher (not significant) in wives vs. husbands during the first 12 months following bereavement.

The range of p-values provided in Figures 2-4 indicates the significance of each of the hazard ratios. For example, in Figure 4, the p-value = 0.0001 associated with the hazard ratio = 1.39 (males; time since bereavement range < 6 months) strongly indicates that the hazard ratio is significantly > 1. Similarly, the p-value = 0.039 associated with the hazard ratio = 1.15 (males; time since bereavement 37-48 months) weakly indicates the significance of the hazard ratio > 1.

Graphical checks of the overall adequacy of the CPH models were performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a reasonable fit to the data (data not shown). The *deviance residual* plots revealed no obvious outliers in the data (data not shown). Further, the Wald test statistic was used to test the fit of the final model,¹⁸ and according to this test statistic, the final model fits the data reasonably well (Supplementary Tables S1, S2, and S3).

DISCUSSION

This is the first comprehensive study to evaluate the relationship between

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3 bereavement and social support in the Amish population. This study provides
4 evidence that Amish widows and widowers have increased mortality risk
5 compared to married cohort members. Although it is difficult to determine
6 whether this effect is of equivalent magnitude as that observed in studies of other
7 populations, the most recent studies of the bereavement effect using the CPH
8 suggest that our findings are generally consistent with data from other
9 populations.⁴

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20 Several previous studies on American, European, and Middle Eastern
21 populations have found that mortality is magnified in individuals widowed at a
22 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
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27 The LDS study is closest to our study because of the large family sizes and the
28 population selected as a religious isolate.⁸ The effects of bereavement on
29 mortality with respect to gender and the age at widowhood ranges observed in the
30 Amish are also largely consistent with those observed in the LDS population,⁸ and
31 other populations such as Finland and Israel (Figure 3).^{6 22} As expected, widowers
32 showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure
33 2).

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44 In the present study, the association between bereavement and mortality is
45 greater in the first 6 months for both men and women (Figure 4), consistent with
46 previous findings.^{2 3 5 20 21 23} The mortality risks in the first 6 months are lower in
47 the Amish (Figure 4) compared to some,^{2 5 20 21} but not all, studies.^{3 23} One
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common pattern observed in this and other studies is that the bereavement effect

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3 is higher in the first 6 months and later life.^{3 23} We did a regression analysis for
4 trend in the data of Figure 4 and there is no significant declining trend after the
5 first 6 months. We speculate that the higher mortality during the first 6 months
6 might reflect acute effects related to the loss of a spouse, while the higher
7 mortality in later life might reflect decreased survival from aging-related diseases
8 that is unmasked in the absence of spousal support.
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11 One strength of our study is that we could evaluate the effect of remarriage
12 because there were substantial numbers of widowed individuals who did and did
13 not get remarried (Table 1) Shor et al. noted that the difficulty in studying the
14 effect of remarriage in populations where all individuals are deceased because “In
15 previous decades, widowed men almost always remarried”.⁴ As suggested by
16 previous studies,^{8 9} remarriage after the death of a spouse significantly influences
17 the bereavement effect because it is associated with increased survival of both
18 males and females across all cohorts . This association is likely influenced by the
19 fact that males or females who get remarried are in sufficiently good physical and
20 mental health to do so. Further, the survival rate is higher for males as compared
21 to females, possibly reflecting support in Amish society for males getting
22 remarried.
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46 Interestingly, more children at the time of the death of the first spouse was
47 associated with increased the risk of death, though the hazard ratio for having > 6
48 surviving children as compared to ≤ 2 was not significantly greater than 1.0. This
49 result does not support the hypothesis that more surviving children confer a
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3 survival advantage to parental longevity, as perhaps by providing social support
4 for their parents. Spouses in the Amish society may also provide unique
5 emotional, psychological, and social support to each other which cannot be
6 provided by their surviving children. The lack of protective association was
7 similarly observed when the number of surviving children was considered as a
8 linear or as a categorical variable (data not shown). This contrasts with data from
9 the Utah Population Database, in which increasing numbers of children were
10 associated with a decreased hazard ratio.⁸ We considered the number of surviving
11 children as a separate term (Table 2a), but did not evaluate the interaction of
12 number of surviving children with widowhood.
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27 The implication of these findings is that differential familial support
28 following bereavement does not appear to be the key factor affecting mortality
29 increases in widowed Amish populations. One potential explanation is that
30 spouses, as “attachment figures,” provide a unique, irreplaceable social support
31 which must be considered independently from other ancillary support providers,
32 such as children.²⁴ Data suggesting that relative mortality risk is also elevated in
33 divorced and never-married populations may support this hypothesis.³ In addition,
34 quality of social support has significantly greater effects on well-being than
35 quantity of support in elderly populations of both sexes.²⁵
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48 In adults aged 65 and older, lower reported social support has been associated
49 with decreased life satisfaction and increased depressive symptoms.²⁶ Family
50 members and close friends should be especially vigilant during the sensitive acute
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3 period following the loss of a spouse, when relative mortality risk is the highest.
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6 There are two limitations of our data. First, the number of surviving children
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8 covariate was simplified because it was treated as a time-independent covariate,
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10 although some children die before their parents, so it could be considered as time-
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12 dependent instead. Second, the causes of death were unavailable. Future research
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14 is needed to study the effect of the loss of a child on the mortality of husbands and
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16 wives by considering the number of surviving children as a time-dependent
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18 covariate. Also, research is needed to determine whether differential measures of
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20 social support may be associated with certain causes of death in widowed
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22 populations. Divorce in the Amish is sufficiently rare that this was not a major
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24 potential source of error.
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30 Our study results indicate that remarriage plays an important role in
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32 improving the survival rate of males and females in the Amish population.
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34 Contrary to results from previous studies, an increase in the number of surviving
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36 children was associated with a decreased survival rate of Amish individuals.
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10
11 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
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13 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
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15 managed the project with assistance from BDM. AAS manages the AGDB project
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17 at the National Institutes of Health and KAR managed the AGDB data at
18
19 University of Maryland Medical School during the period of this project. ARS
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21 provided resources and advice. All authors proofread and approved the
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23 manuscript.
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37 Medical Center.
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42 **Competing interests** None.
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46 **Ethics approval** The construction and maintenance of AGDB are covered under
47
48 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
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50 Investigator.
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54 **Provenance and peer review** Not commissioned; externally peer reviewed.
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FIGURE LEGENDS

Figure 1. A flow diagram that represents all the steps performed for filtering 10,892 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. “Unknown spouse” refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. “Birth year too late” means that the birth year of the husband or wife is known and is > 1901 . “Dates not recognized by R” are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. “Implausible birth or death dates” refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a); * and ** on the top of the blocks represent significance of hazard ratios with $p\text{-value} < 0.05$ and $p\text{-value} < 0.001$, respectively.

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood (design provided in Table 2b); NSC1: Number of Surviving Children (3-6 vs ≤ 2); NSC2: Number of Surviving

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3 Children (>6 vs ≤ 2); * and ** on the top of the blocks represent significance of
4 hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.
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8 Figure 4. Hazard ratios of widowed husbands and wives versus their married
9 counterparts according to time since bereavement (months) (design provided in
10 Table 2c); * and ** on the top of the blocks represent significance of hazard ratios
11 with p-value < 0.05 and p-value < 0.001, respectively.
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TABLES

Table 1: Characteristics of 10,892 spouse pairs according to birth cohort of husband.

Cohorts	Pre-1850	1850-1875	1876-1900
Number of couples	3711	3210	3971
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2043(51.4)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	1928(48.2)
Mean husband age at widowhood	63.0	59.8	62.2
Mean wife age at widowhood	62.0	62.8	66.9
Mean widowed husband survival in years	14.4	18.3	18.4
Mean widowed wife survival in years	15.3	16.0	16.6
Mean husband age at death	71.1	72.0	74.6
Mean wife age at death	69.6	68.5	72.6
Mean age difference husband-wife	3.4	2.8	2.0
Mean number of children	5.4	5.3	5.3
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	648(33.6)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	180(8.8)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0
ID ₁	51	52	0	0	0	0	0	0	1	0

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ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

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Analysis of the Bereavement Effect After the Death of a Spouse in the Amish

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ABSTRACT

Objective This study investigates the association between bereavement and the mortality of a surviving spouse among Amish couples. We hypothesized that the bereavement effect would be relatively small in the Amish due to the unusually cohesive social structure of the Amish that might attenuate the loss of spousal support.

Design Population-based cohort study

Setting United States of America

Participants 10,892 Amish couples born during 1725-1900 located in Pennsylvania, Ohio, and Indiana. All the participants are deceased.

Outcome measure The survival time is 'age'; event is 'death'. Hazard ratios of widowed individuals with respect to gender, age at widowhood, remarriage, number of surviving children, and time since bereavement.

Results We observed hazard ratios for widowhood ranging from 1.06-1.26 over the study period (nearly all differences significant at $p < 0.05$). Mortality risks tended to be higher in men than in women and in younger compared to older bereaved spouses. There were significantly increased mortality risks in both widows and widowers who did not remarry. We observed a higher number of surviving children to be associated with increased mortality in both males and females. Mortality risk following bereavement was higher in the first 6 months among both men and women.

Conclusions We conclude that bereavement effects remain apparent even in this

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9 socially cohesive Amish community. Remarriage is associated with a significant
10 decrease in the mortality risk among Amish individuals. Contrary to results from
11 previous studies, an increase in the number of surviving children was associated
12 with decreased survival rate.
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ARTICLE SUMMARY

Article focus

- The focus of this article is to evaluate the relationship between bereavement and the mortality of a surviving spouse in the Amish population.~~The focus of this article is to evaluate the relationship between bereavement and social support in the Amish population.~~
- We evaluated the association of bereavement and mortality of a surviving spouse with respect to gender, age at widowhood, and time since bereavement while accounting for remarriage, and number of surviving children using Cox proportional hazard models.~~We evaluated the association of surviving spouse gender, age at widowhood, remarriage, number of surviving children, and time since bereavement on the bereavement effect using Cox proportional hazard models.~~

Key messages

- This study shows that strong social support is not sufficient to decrease the mortality risks among Amish individuals, and therefore the support provided by the spouse is irreplaceable.
- Remarriage significantly decreases the mortality risk for both males and females.

Strengths and limitations of this study

- Due to the availability of remarriage status of the surviving spouses, we could reproduce and quantify the decreased bereavement effect associated with remarriage.
- Due to the unavailability of data on causes of death, it was not possible to determine the relationship between differential measures of social support and certain causes of death.
- The number of surviving children was simplified to be a time independent covariate although it may change between the death of the first parent and the death of the second parent.

INTRODUCTION

It has been consistently established that widowed individuals exhibit increased mortality compared to married individuals.¹⁻⁴ This excess mortality risk, referred to as the “bereavement effect,” is strongest in the first few years following widowhood, among men who outlive their wives, and among younger widows/widowers.²⁻⁵ Factors proposed to explain this phenomenon include: the acute grief and stress of bereavement; shared environmental risk factors between spouses; marital selection; economic hardship; and loss of social support.^{6,7}

These influences are difficult to isolate and independently assess. Several studies have shown that larger social network size and greater level of social support are generally associated with lower rates of mortality and morbidity.⁴ In a study based on the Utah Population Database, mortality risk among widowers decreased with membership in the Church of Latter Day Saints (LDS, a.k.a. Mormon Church) and with increasing numbers of children. These factors were interpreted as proxies for social support.⁸ Additionally, a study in Washington County, Maryland, indicated that living alone was a risk factor for increased mortality in widowed populations.⁹ Loss of social support following the death of a spouse is generally more profound in widowers than in widows, but there is currently no empirical evidence that this difference mediates the higher mortality consistently observed among widowed husbands compared to widowed wives.¹⁰

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The Amish maintain a cultural identity distinct from mainstream American

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9 culture that is characterized by their traditional dress, a plain lifestyle, and non-
10 adoption of modern technology (e.g., electricity, cars, telephones), German
11 dialect, separate school system, and ultra-conservative Anabaptist religious
12 practices. A central tenet of Amish culture throughout their history in the USA
13 has been social cohesiveness with emphasis on family and community. Members
14 of this tight-knit society have extraordinary social support from cradle to grave,
15 including community-financing of medical costs and support during times of
16 need. The community financing of medical costs became increasingly formalized
17 during the 20th century to the point where it is now a formal system of self-
18 insurance called “Amish Aid”.
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22 To gain a different perspective on potential influences of social support on
23 bereavement, we assessed the bereavement effect in an Amish population, whose
24 culture is characterized by its core beliefs in community and social cohesion.
25 Examples of the strong degree of social support within the Amish include
26 community-managed health insurance and community ties through membership in
27 the church, and the sharing of ultraconservative cultural beliefs and lifestyles.¹²
28 We reasoned that the high level of social support in the Amish population might
29 mitigate the bereavement effect. Previous studies used covariates such as
30 education (homogeneous in the Amish),¹² health habits, remarriage, church visits,
31 neighborhood interaction, and household size to study the relationship between
32 bereavement and social support.^{4 8 13}
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48 We estimated bereavement effects in widows and widowers separately, and
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9 further assessed mortality risks as a function of age at widowhood and time since
10 bereavement.^{2 8} We used Cox proportional hazard (CPH) models to analyze the
11 association of bereavement and mortality of widowed husbands and wives. We
12 considered remarriage and number of surviving children as additional potential
13 modifiers of the bereavement effect.
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20 MATERIAL AND METHODS

21 Data source

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23 The Anabaptist Genealogy Database (AGDB) version 5 was used to study the
24 effect of bereavement on the Amish population.^{14 15} The AGDB is a
25 computerized genealogy database of Amish and some Mennonite populations, last
26 updated in 2010. This database includes information about the family
27 relationships and the birth, marriage, and death dates of children of Amish
28 individuals from North America, mostly located in Pennsylvania, Ohio, and
29 Indiana. The "individual table" of AGDB contains information about 539,822
30 individuals. The "relationship table" includes information about 136,213 Amish
31 couples. An individual who is married multiple times participates in multiple
32 relationship table entries. There are 1,369 relationship entries among the 136,213
33 entries concerning children for whom one or both biological parents are unknown
34 (Figure 1).
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46 Amish people rarely give birth out of wedlock, and a vast majority of the
47 mating samples we used are documented as marriages in the three original sources
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9 from which AGDB is derived. However, there is less availability of marriage
10 dates than birth dates; we did not wish to exclude couples with missing marriage
11 dates. For this study, we included couples in which both spouses met four
12 conditions: (1) they were born prior to January 1, 1901; (2) their birth and death
13 dates were recorded; (3) they were in their first marriage; and (4) they did not die
14 on the same day. Constraint (4) excludes couples who may have died from a
15 shared disaster (e.g., accident). A total of 10,892 couples met the inclusion
16 criteria. The oldest person included was born on February 14, 1725, and the
17 youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were
18 applied.
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22 Our data set included 10,892 couples with information on their date of birth,
23 date of death, number of surviving children, family IDs, and remarriage status
24 after widowhood. For some analyses, these couples were first partitioned into
25 three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and
26 based on both husband's and wife's birth year (1876-1900) and sex. For the time
27 since bereavement analyses, we did not partition into cohorts because the number
28 of widow(er)s who died within 6 months after being bereaved is small (N=291).
29 Characteristics of these couples are shown in Table 1.
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31

32 **Statistical analyses**

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34 Similar to several past analyses,^{2 8 13 16} we used the Cox Proportional
35 Hazards (CPH) model where the response variable or survival time is 'age at
36 death' and the event is 'death'. The model is used to study the association of
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9 widowhood and mortality rates in the surviving spouse, while adjusting for
10 covariates such as education, health habits, age in years, number of children,
11 and remarriage. In some of our analyses, we adjusted for remarriage and
12 number of children as covariates; we did not adjust for education or health
13 habits. Husbands and wives were always analyzed separately.
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18 The main covariate, widowhood, was monitored as a time-dependent
19 covariate, by assigning a value of 1 for each time period the individual was
20 widowed and 0 otherwise. The remarriage covariate was also a time-dependent
21 covariate, by assigning a value 1 for the widowed period if the individual
22 remarried and 0 otherwise. The number of surviving children was a time-
23 independent covariate and counted at the beginning of widowhood period of the
24 surviving spouse. The covariate was divided into three categories, as follows:
25 0=number of surviving children is ≤ 2 , 1=number of surviving children is between
26 3 and 6, and 2=number of surviving children is >6 . The categories ≤ 2 children, 3-
27 6 children, and > 6 children are separate. These boundaries were chosen to ensure
28 categories that were roughly balanced in size. To evaluate any possible bias by
29 counting the number of surviving children at the death of the first spouse, we also
30 estimated how many couples changed categories between the death of the first
31 spouse and the death of second spouse due to death of one or more children
32 during the widowhood period.
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46 Survival analyses were performed using the R programming environment.¹⁷
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48 All estimates and confidence intervals were obtained using the `coxph` function
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9 available in the `survival` package.

10 11 **Representation of survival data for CPH analysis**

12 We show below examples of how we represented the survival data for CPH
13 analysis using a time modeling approach. To represent the survival data columns, the
14 standard approach is to convert the couple's demographic information, date of
15 death, date of birth, remarriage and number of surviving children into columns
16 representing **start** time, **stop** time, **event** status, and all of the included covariates
17 (widowhood, remarriage, and number of surviving children).^{18 19} Here (**start**,
18 **stop**) is an interval of risk, open on the left and closed on the right; the **event**
19 column is set to 1 if the subject had an event (death) at time **stop** and is 0
20 otherwise. To illustrate the subtleties, we consider a hypothetical example of
21 three wives (ID₁, ID₂ and ID₃) from the cohort 1850-1875.
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33 • *ID*₁: born on 01/01/1860; widowed on 01/01/1907 at age 47; got remarried;
34 number of surviving children = 3 and eventually died on 01/01/1923 at age 63.
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39 • *ID*₂: born on 09/01/1870; widowed on 09/01/1930 at age 60; never got
40 remarried; number of surviving children = 6 and died on 09/01/1955 at age 85.
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45 • *ID*₃: born on 03/01/1871; never widowed and never remarried; number of
46 surviving children = 4 and died on 04/01/1930 at age 59.
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9 The data used for the CPH considering widowhood as a single time-
10 dependent covariate is presented in Table 2a. The following model was used to
11 estimate the association between widowhood and mortality.
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$$13 \quad h(t) = h_0(t) \exp[\beta W]$$

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16 This model can be run with additional covariates and results that adjust for the
17 ability to remarry and the number of surviving children are presented.
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20 **Timing of widowhood**

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22 The above model considers widowhood as a single time-dependent
23 covariate, evaluating the association between mortality and being
24 widowed. One can further ask if that association varies over the life
25 course by considering the impact of age at widowhood on mortality.
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27 We addressed this issue following the approach of Mineau et al. by
28 adding terms for age at widowhood into the model as covariates (<45,
29 45-54, 55-64, 65-74 and >75) to allow the hazard ratio (HR) to vary according to
30 age at widowhood (see Table 2b).⁸ These terms were included in the model
31 as time-dependent dummy variables, created to represent the
32 widowhood experience of each individual across the 5 age windows
33 spanning the individual's age: $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , and W_{75+} .
34
35 Further, we also addressed the impact of remarriage and number of
36 surviving children on the mortality of Amish individuals.
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40 The following model can be used to estimate the association
41 between each of the five dummy variables and mortality:
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$$h(t) = h_0(t) \exp[\sum \beta_j W_j + \beta_6 R + \beta_7 C] \text{ with } j = 1, \dots, 5$$

where W_1, \dots, W_5 dummy variables are associated with $W_{<45}$, W_{45-54} , W_{55-64} , W_{65-74} , W_{75+} columns provided in the Table 2b.

Time since bereavement

Next, we evaluated the association between mortality and time since bereavement or widowhood. We followed the approach of Schaefer et al. by considering the following time since bereavement ranges: 0-6 months, 7-12 months, 13-24 months, 25-36 months, 37-48 months, 49-60 months and > 60 months.² This approach allowed us to estimate HR according to time since bereavement (see Table 2c). The columns TSB₀₋₆, TSB₇₋₁₂, ... in Table 2c are time dependent covariates that change with the survival time associated with widowed husbands and wives. We did not account for remarriage in this analysis because of missing remarriage dates.

The following model was used to study the association between mortality and time since bereavement:

$$h(t) = h_0(t) \exp[\sum \beta_j W_j] \text{ with } j = 1, \dots, 7$$

where W_1, \dots, W_7 dummy variables are associated with the TSB₀₋₆, TSB₇₋₁₂, TSB₁₃₋₂₄, TSB₂₅₋₃₆, TSB₃₇₋₄₈, TSB₄₉₋₆₀ and TSB_{>60} columns provided in the Table 2c.

RESULTS

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9 We initially partitioned the 10,892 couples from AGDB into three cohorts ranging
10 in size from 3,210 (husband birth year 1850-1875) to 3,971 (husband birth year
11 1876-1900). Wives were more likely to die after their husbands in all three
12 cohorts while husbands had a higher mean age at death than their wives. The
13 average age differences between the husbands and wives for all cohorts are shown
14 in Table 1. The number of remarried wives was far smaller than the number of
15 remarried husbands (n = 1,290 widowed husbands vs 378 widowed wives). The
16 number and proportion of remarried husbands and wives were increased in the
17 more contemporary cohorts (see Table 1). In contrast to other populations from
18 the 19th and early 20th centuries,⁴ the majority of widowed husbands did not
19 remarry, making it interesting to study the effect of remarriage in the Amish
20 population.
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31 **Hazard Ratios**

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33 The overall hazard ratios (HR), and their 95% confidence intervals (CIs),
34 associated with widowhood are displayed in Figure 2 according to birth cohort.
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36 The significance of these hazard ratios are indicated with the help of p-values
37 (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2.
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39 These hazard ratios were estimated using the CPH model and design provided in
40 Table 2a. As expected, widowhood was associated with increased mortality for
41 both husbands and wives following the death of spouse, with HRs ranging
42 from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed
43 husbands in cohort 1850-1875). The impact of widowhood was
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9 disproportionately greater on surviving husbands than on surviving wives in the
10 pre-1850 and 1876-1900 cohorts, although there was little difference in mortality
11 between widowed husbands vs. wives in the 1850-1875. There was no clear trend
12 showing that the hazard ratios changed across cohorts in either surviving
13 husbands or wives.
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18 Figure 3 shows the hazard ratios along with 95% CIs, and range of p-values
19 for the model in which age at widowhood is accounted for (design model in Table
20 2b). These results reveal markedly higher bereavement effects in each age at
21 widowhood group compared to the results shown in Figure 2, consistent with the
22 notion that the bereavement effect may be diminished over time and most
23 pronounced in the years proximal to the death of the spouse.^{2 5 20 21} In nearly all
24 age at widowhood categories, the bereavement effect is stronger in widowed
25 husbands than in widowed wives.
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33 The number of children surviving when the first spouse died (with two
34 pairwise comparisons 3-6 vs ≤ 2 ; > 6 vs ≤ 2) was included as a time-independent
35 covariate in the models whose results are shown in Figure 3. In general, there
36 was a very weak association between number of surviving children and mortality
37 in husbands and wives. Contrary to our expectations and a prior study,⁸ in each
38 case, a higher number of surviving children was not significantly associated with
39 lower mortality in both husbands and wives (See Figure 3). Further, the results in
40 Figure 3 show that the effect of bereavement decreases if the Amish individual
41 remarries.
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9 There are two potential sources of bias in analyses involving the number of
10 surviving children covariate. Couples in which one spouse died at age <50 are
11 likely not to have as many children as couples in which both parents survived to
12 at least age 50. To quantify this bias, we repeated the analysis in Figure 3 by
13 excluding all the couples who got widowed before age 50 (data not shown). The
14 results show that there is no significant change in the hazard ratios vs. Figure 3.
15 The maximum change for any of the hazard ratios related to the number of
16 surviving children was 0.03 (data not shown).
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24 On the other end of the age spectrum, there is a potential source of bias as
25 more children may die, the longer the surviving parent lives. Because the number
26 of children was divided into three categories ≤ 2 , 3-6, and > 6 , any possible bias
27 could arise when and only when the death of a child shifted the number of
28 surviving children from a higher category to a lower category. Over all couples,
29 the number of widows or widowers whose number of surviving children changed
30 to a lower category from the date of widowhood to the date of death was only 298
31 (< 3% of all couples).
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39 When the analyses corresponding to Figure 3 were repeated excluding these
40 298 couples, the hazard ratios were essentially unchanged. The maximum change
41 was 0.04; sometimes the hazard ratio increased slightly and sometime it decreased
42 slightly (data not shown).
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46 Hazard ratios and 95% CIs for the time since bereavement analysis are shown
47 in Figure 4. The significant p-values are indicated in Figure 4. These results were
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obtained using the CPH model and the design defined in Table 2c. The results show that there is a high risk of mortality for recently widowed husbands and wives. Further, the hazard is higher (not significant) in wives vs. husbands during the first 12 months following bereavement. Hazard ratios and 95% CIs for the time since bereavement analysis are shown in Figure 4. The significant p values are indicated in Figure 4. These results were obtained using the CPH model and the design defined in Table 2c. The results show that there is an increase in risk of mortality for recently widowed husbands and wives, and the hazard decreases with time since bereavement but remains significantly greater than 1. Further, the hazard is higher (not significant) in wives vs. husbands during the first 12 months following bereavement.

The range of p-values provided in Figures 2-4 indicates the significance of each of the hazard ratios. For example, in Figure 4, the p-value = 0.0001 associated with the hazard ratio = 1.39 (males; time since bereavement range < 6 months) strongly indicates that the hazard ratio is significantly > 1. Similarly, the p-value = 0.039 associated with the hazard ratio = 1.15 (males; time since bereavement 37-48 months) weakly indicates the significance of the hazard ratio > 1.

Graphical checks of the overall adequacy of the CPH models were performed.^{18 19} Based on the *Cox-Snell residuals* plot, the final model gave a reasonable fit to the data (data not shown). The *deviance residual* plots revealed no obvious outliers in the data (data not shown). Further, the Wald test

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9 statistic was used to test the fit of the final model,¹⁸ and according to this test
10 statistic, the final model fits the data reasonably well (Supplementary Tables S1,
11 S2, and S3).
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14 15 16 **DISCUSSION** 17

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20 This is the first comprehensive study to evaluate the relationship between
21 bereavement and social support in the Amish population. This study provides
22 evidence that Amish widows and widowers have increased mortality risk
23 compared to married cohort members. Although it is difficult to determine
24 whether this effect is of equivalent magnitude as that observed in studies of other
25 populations, the most recent studies of the bereavement effect using the CPH
26 suggest that our findings are generally consistent with data from other
27 populations.⁴
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31 Several previous studies on American, European, and Middle Eastern
32 populations have found that mortality is magnified in individuals widowed at a
33 younger age and that widowers have higher mortality risk than widows.^{3 5 8 16 20 22}
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35 The LDS study is closest to our study because of the large family sizes and the
36 population selected as a religious isolate.⁸ The effects of bereavement on
37 mortality with respect to gender and the age at widowhood ranges observed in the
38 Amish are also largely consistent with those observed in the LDS population,⁸ and
39 other populations such as Finland and Israel (Figure 3).^{6 22} As expected, widowers
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showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure 2).

~~In the present study, the association between bereavement and mortality is greater in the first 6 months for both men and women (Figure 4), consistent with previous findings.^{2,3,5,20,21,23} The mortality risks in the first 6 months are lower in the Amish (Figure 4) compared to some studies,^{2,5,20,21} but not all studies.^{3,23} One common pattern observed in this and other studies is that the initially high bereavement effect first decreases but then increases with time since bereavement.³⁻²³ We speculate that the increased mortality during the first 6 months might reflect acute effects related to the loss of a spouse, while the gradual increases in mortality emerging in later life might reflect decreased survival from aging-related diseases that is unmasked in the absence of spousal support.~~ In the present study, the association between bereavement and mortality is greater in the first 6 months for both men and women (Figure 4), consistent with previous findings.^{2,3,5,20,21,23} The mortality risks in the first 6 months are lower in the Amish (Figure 4) compared to some^{2,5,20,21} but not all, studies.^{3,23} One common pattern observed in this and other studies is that the bereavement effect is higher in the first 6 months and later life.^{3,23} We did a regression analysis for trend in the data of Figure 4 and there is no significant declining trend after the first 6 months. We speculate that the higher mortality during the first 6 months might reflect acute effects related to the loss of a spouse, while the higher mortality in later life might reflect decreased survival from aging-related diseases

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9 that is unmasked in the absence of spousal support.

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11 One strength of our study is that we could evaluate the effect of remarriage
12 because there were substantial numbers of widowed individuals who did and did
13 not get remarried (Table 1) Shor et al. noted that the difficulty in studying the
14 effect of remarriage in populations where all individuals are deceased because “In
15 previous decades, widowed men almost always remarried”.⁴ As suggested by
16 previous studies,^{8,9} remarriage after the death of a spouse significantly influences
17 the bereavement effect because it is associated with increased survival of both
18 males and females across all cohorts. This association is likely influenced by the
19 fact that males or females who get remarried are in sufficiently good physical and
20 mental health to do so. Further, the survival rate is higher for males as compared
21 to females, possibly reflecting support in Amish society for males getting
22 remarried.
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33 Interestingly, more children at the time of the death of the first spouse was
34 associated with increased the risk of death, though the hazard ratio for having > 6
35 surviving children as compared to ≤ 2 was not significantly greater than 1.0. This
36 result does not support the hypothesis that more surviving children confer a
37 survival advantage to parental longevity, as perhaps by providing social support
38 for their parents. Spouses in the Amish society may also provide unique
39 emotional, psychological, and social support to each other which cannot be
40 provided by their surviving children. The lack of protective association was
41 similarly observed when the number of surviving children was considered as a
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9 linear or as a categorical variable (data not shown). This contrasts with data from
10 the Utah Population Database, in which increasing numbers of children were
11 associated with a decreased hazard ratio.⁸ We considered the number of surviving
12 children as a separate term (Table 2a), but did not evaluate the interaction of
13 number of surviving children with widowhood. Interestingly, increasing numbers
14 of surviving children at the time of widowhood did not confer a survival
15 advantage for Amish individuals. This result was counter to our hypothesis that
16 children can help provide social support for their parents. The hazard ratio was
17 greater than 1.0 (but not significant) for all Amish individuals with number of
18 surviving children > 6 as compared to ≤ 2. Spouses in the Amish society may also
19 provide unique emotional, psychological, and social support to each other which
20 cannot be provided by their surviving children. The lack of protective association
21 was similarly observed when the number of surviving children was considered as
22 a linear or as a categorical variable (data not shown). This contrasts with data
23 from the Utah Population Database, in which increasing numbers of children were
24 associated with a decreased hazard ratio.⁸

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39 The implication of these findings is that differential familial support
40 following bereavement does not appear to be the key factor affecting mortality
41 increases in widowed Amish populations. One potential explanation is that
42 spouses, as “attachment figures,” provide a unique, irreplaceable social support
43 which must be considered independently from other ancillary support providers,
44 such as children.²⁴ Data suggesting that relative mortality risk is also elevated in

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9 divorced and never-married populations may support this hypothesis.³ In addition,
10 quality of social support has significantly greater effects on well-being than
11 quantity of support in elderly populations of both sexes.²⁵
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14 In adults aged 65 and older, lower reported social support has been associated
15 with decreased life satisfaction and increased depressive symptoms.²⁶ Family
16 members and close friends should be especially vigilant during the sensitive acute
17 period following the loss of a spouse, when relative mortality risk is the highest.
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21 There are two limitations of our data. First, the number of surviving children
22 covariate was simplified because it was treated as a time-independent covariate,
23 although some children die before their parents, so it could be considered as time-
24 dependent instead. Second, the causes of death were unavailable. Future research
25 is needed to study the effect of the loss of a child on the mortality of husbands and
26 wives by considering the number of surviving children as a time-dependent
27 covariate. Also, research is needed to determine whether differential measures of
28 social support may be associated with certain causes of death in widowed
29 populations. Divorce in the Amish is sufficiently rare that this was not a major
30 potential source of error.
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34 Our study results indicate that remarriage plays an important role in
35 improving the survival rate of males and females in the Amish population.
36 Contrary to results from previous studies, an increase in the number of surviving
37 children was associated with a decreased survival rate of Amish individuals.
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41 **Acknowledgments** Thanks to two reviewers for excellent suggestions that led us
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9 to improve the manuscript.

10
11 **Contributors** BDM conceived the study. AS and SS did the research and data
12 analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS
13 wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS
14 managed the project with assistance from BDM. AAS manages the AGDB project
15 at the National Institutes of Health and KAR managed the AGDB data at
16 University of Maryland Medical School during the period of this project. ARS
17 provided resources and advice. All authors proofread and approved the
18 manuscript.
19

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26 Medical Center.
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29 **Competing interests** None.
30

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32 **Ethics approval** The construction and maintenance of AGDB are covered under
33 an IRB-approved human subjects protocol at the NIH, Leslie Biesecker, Principal
34 Investigator.
35

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37 **Provenance and peer review** Not commissioned; externally peer reviewed.
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9 **Data sharing statement** Data from the AGDB are available to investigators
10 (including ARS and various others not participating in this study) who have an
11 IRB-approved protocol to study the Amish or other Anabaptist groups.
12 Investigators can request access to AGDB by writing to AAS and to Dr.
13 Biesecker.
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FIGURE LEGENDS

Figure 1. A flow diagram that represents all the steps performed for filtering 10,892 couples from total 136,213 couples available in AGDB. In the flow diagram, each couple is counted as excluded only once, even if multiple exclusion criteria apply. “Unknown spouse” refers to entries in the AGDB relationship table in which at least one parent is unknown; almost all of these entries are for adopted children for whom at least one of the biological parents is unknown. Because AGDB is used primarily in genetic studies (unlike this study), the distinction between biological and adoptive relationships is stored. “Birth year too late” means that the birth year of the husband or wife is known and is > 1901 . “Dates not recognized by R” are invalid dates such as the 31st of June, which got into AGDB due to errors in the original sources. “Implausible birth or death dates” refers to a few individuals who are shown as married but have lifespans of less than 10 years likely due to typos in the birth year in the original sources.

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a); * and ** on the top of the blocks represent significance of hazard ratios with $p\text{-value} < 0.05$ and $p\text{-value} < 0.001$, respectively.

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood (design provided in Table 2b); NSC1: Number of Surviving Children (3-6 vs ≤ 2); NSC2: Number of Surviving

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9 Children (>6 vs ≤ 2); * and ** on the top of the blocks represent significance of
10 hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.

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12 Figure 4. Hazard ratios of widowed husbands and wives versus their married
13 counterparts according to time since bereavement (months) (design provided in
14 Table 2c); * and ** on the top of the blocks represent significance of hazard ratios
15 with p-value < 0.05 and p-value < 0.001, respectively.
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TABLES

Table 1: Characteristics of 10,892 spouse pairs according to birth cohort of husband.

Cohorts	Pre-1850	1850-1875	1876-1900
Number of couples	3711	3210	3971
Number (%) of wives out surviving their husband	2043(55.0)	1661(51.7)	2043(51.4)
Number (%) of husbands out surviving their wife	1668(45.0)	1549(48.3)	1928(48.2)
Mean husband age at widowhood	63.0	59.8	62.2
Mean wife age at widowhood	62.0	62.8	66.9
Mean widowed husband survival in years	14.4	18.3	18.4
Mean widowed wife survival in years	15.3	16.0	16.6
Mean husband age at death	71.1	72.0	74.6
Mean wife age at death	69.6	68.5	72.6
Mean age difference husband-wife	3.4	2.8	2.0
Mean number of children	5.4	5.3	5.3
Number (%) of widowed husbands remarried	238(14.2)	404(26.0)	648(33.6)
Number (%) of widowed wives remarried	58(2.8)	140(8.4)	180(8.8)

Table 2a-c. The three tables below illustrate encodings for different CPH designs to test for the bereavement effect, following the general syntax of CPH table designs recommended in (18). See example description in Material and Methods.

Table 2a: Data structure for Cox Proportional Hazard model that does not estimate the effect for different ages, but instead estimates only widowed vs. non-widowed.

ID	start	stop	event	W	R	C
1	0	47(age at widowhood)	0	0	0	3
1	47	63(age at death)	1	1	1	3
2	0	60(age at widowhood)	0	0	0	6
2	60	85(age at death)	1	1	0	6
3	0	59(age at death)	1	0	0	4

Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

ID	start	stop	event	W _{<45}	W ₄₅₋₅₄	W ₅₅₋₆₄	W ₆₅₋₇₄	W ₇₅₊	R	C
1	0	47	0	0	0	0	0	0	0	3
1	47	63	1	0	1	0	0	0	1	3
2	0	60	0	0	0	0	0	0	0	6
2	60	85	1	0	0	1	0	0	0	6
3	0	59	1	0	0	0	0	0	0	4

Table 2c: Data structure for Cox Proportional Hazard model that estimates association between widowhood with respect to time since bereavement and mortality. Start and Stop columns are in years and Time Since Bereavement (TSB) columns are in months.

ID	start	stop	event	TSB ₀₋₆	TSB ₇₋₁₂	TSB ₁₃₋₂₄	TSB ₂₅₋₃₆	TSB ₃₇₋₄₈	TSB ₄₉₋₆₀	TSB _{>60}
ID ₁	0	47	0	0	0	0	0	0	0	0
ID ₁	47	47.5	0	1	0	0	0	0	0	0
ID ₁	47.5	48	0	0	1	0	0	0	0	0
ID ₁	48	49	0	0	0	1	0	0	0	0
ID ₁	49	50	0	0	0	0	1	0	0	0
ID ₁	50	51	0	0	0	0	0	1	0	0
ID ₁	51	52	0	0	0	0	0	0	1	0

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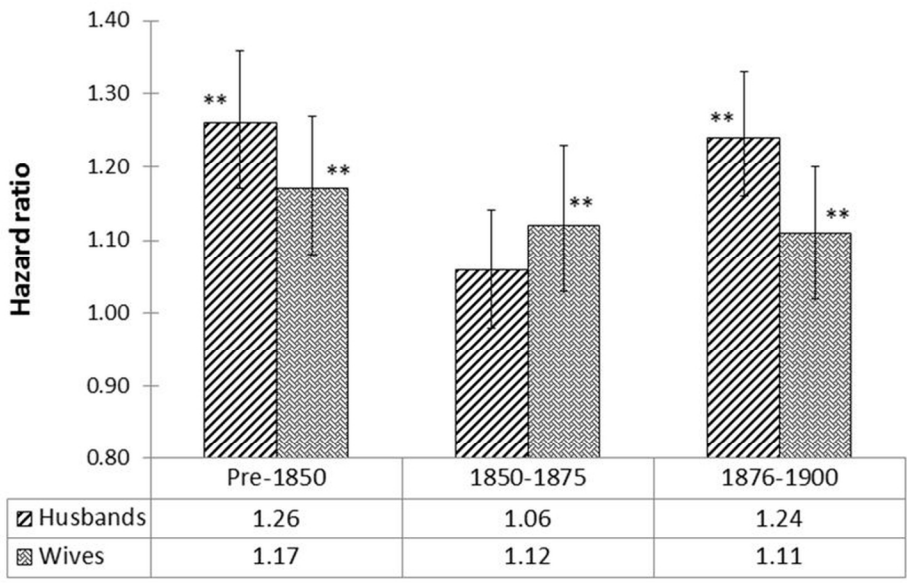
ID ₁	52	63	1	0	0	0	0	0	0	1
ID ₂	0	60	0	0	0	0	0	0	0	0
ID ₂	60	60.5	0	1	0	0	0	0	0	0
ID ₂	60.5	61	0	0	1	0	0	0	0	0
ID ₂	61	62	0	0	0	1	0	0	0	0
ID ₂	62	63	0	0	0	0	1	0	0	0
ID ₂	63	64	0	0	0	0	0	1	0	0
ID ₂	64	65	0	0	0	0	0	0	1	0
ID ₂	65	85	1	0	0	0	0	0	0	1
ID ₃	0	59	1	0	0	0	0	0	0	0

For peer review only



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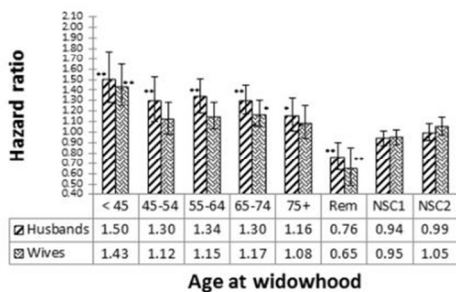


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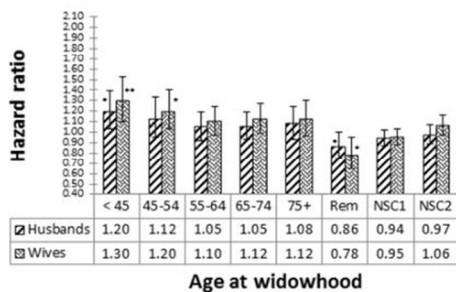
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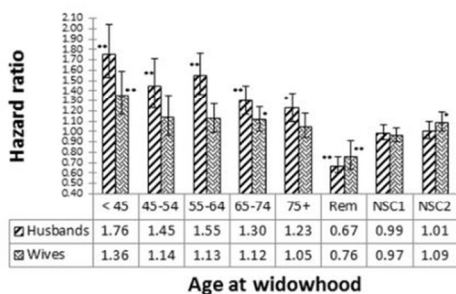
Cohort: Pre-1850



Cohort: 1850-1875



Cohort: 1876-1900



123x90mm (300 x 300 DPI)

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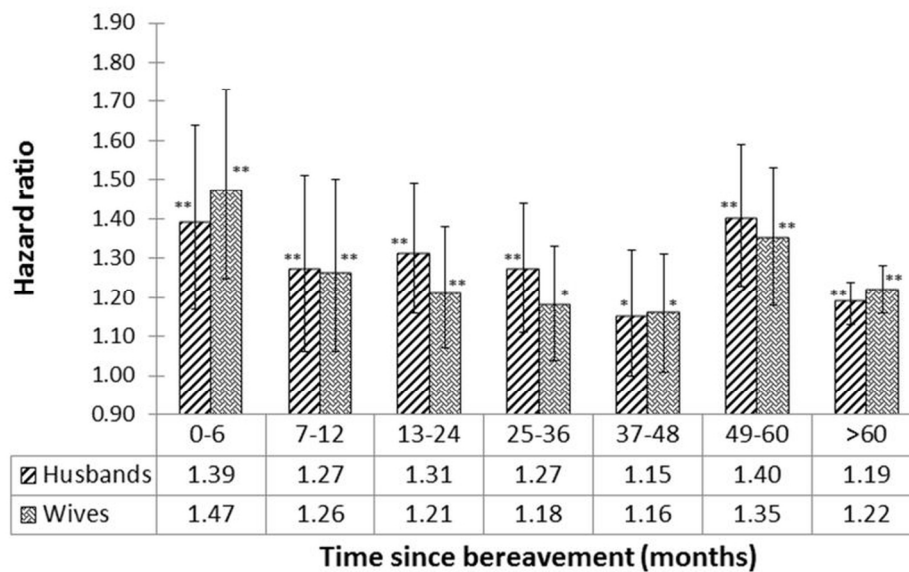


Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months) (design provided in Table 2c); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.

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Table S1: Wald statistics associated with models that are used to study the association between widowhood and mortality.

Cohorts	Pre-1850	1850-1875	1876-1900
Husbands	38.2 ^{**}	5.9 [*]	38.0 ^{**}
Wives	15.2 ^{**}	6.9 [*]	6.8 [*]
* P<0.05 and ** P<0.001			

Table S2: Wald statistics associated with models that are used to study the association between timing of widowhood and mortality.

Cohorts	Pre-1850	1850-1875	1876-1900
Husbands	57.2 ^{**}	15.8 [*]	87.7 ^{**}
Wives	39.8 ^{**}	20.3 ^{**}	28.9 ^{**}
* P<0.05 and ** P<0.001			

Table S3: Wald statistics associated with models that are used to study the association between time since bereavement and mortality.

Husbands	94.6 ^{**}
Wives	84.3 ^{**}
** P<0.001	