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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

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.

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.

- 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.

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 11]

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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.

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. 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. 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 analyses

Similar to several past analyses,[2 8 13 16] we used the Cox Proportional Hazards (CPH) model 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.

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 divided into 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.

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

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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 (ID_1, ID_2 and ID_3) 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:

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

where $W_1,\ldots,W_5$ 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:

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

where $W_1,\ldots,W_7$ 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

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

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

Hazard Ratios

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

Figure 2 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 1, 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 2. 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. Further, the results in Figure 2 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 3. 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.

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. Further, the *deviance residual* plots showed that there were no obvious outliers in the data.

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 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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).

In the present study, the association between bereavement and mortality is greater in the first 6 months for both men and women (Figure 3), consistent with previous findings.[2 3 5 20 21 23] The mortality risks in the first 6 months are lower in the Amish (Figure 3) 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.

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). Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because “In previous decades, widowed men almost always remarried".[4] 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.

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. The lack of protective association was similarly observed when the number of surviving children was considered as a linear or as a categorical variable (data not shown). This contrasts with data from the Utah Population Database, in which increasing numbers of children were associated with a decreased hazard ratio.[8]

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

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

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.

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.

Contributors BDM conceived the study. AS and SS did the research and data analysis, with substantial guidance from PFM, BDM, and AAS. AS and SS wrote the manuscript, with substantial editing by PFM, BDM, and AAS. AAS managed the project with assistance from BDM. AAS manages the AGDB project at the National Institutes of Health and KAR managed the AGDB data at

University of Maryland Medical School during the period of this project. ARS provided resources and advice. All authors proofread and approved the manuscript.

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Competing interests None.

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

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups. Investigators can request access to AGDB by writing to AAS and to Dr. 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

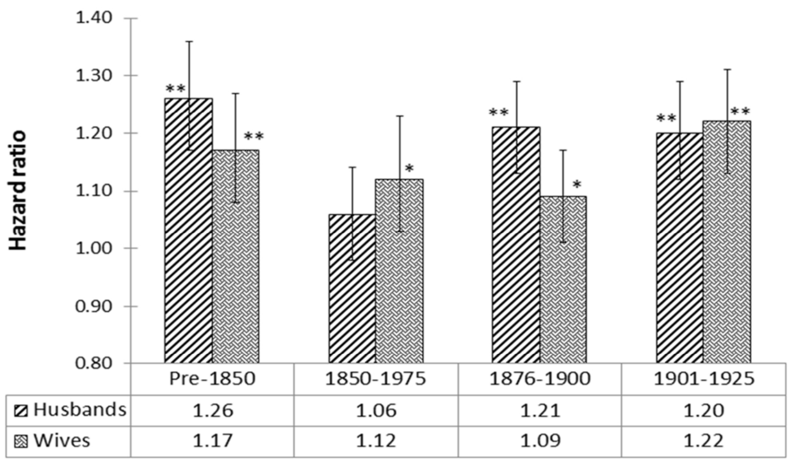

Figure 1. Hazard ratios of widowed husbands and wives versus their married counterparts (design provided in Table 2a).
173x233mm (300 x 300 DPI)

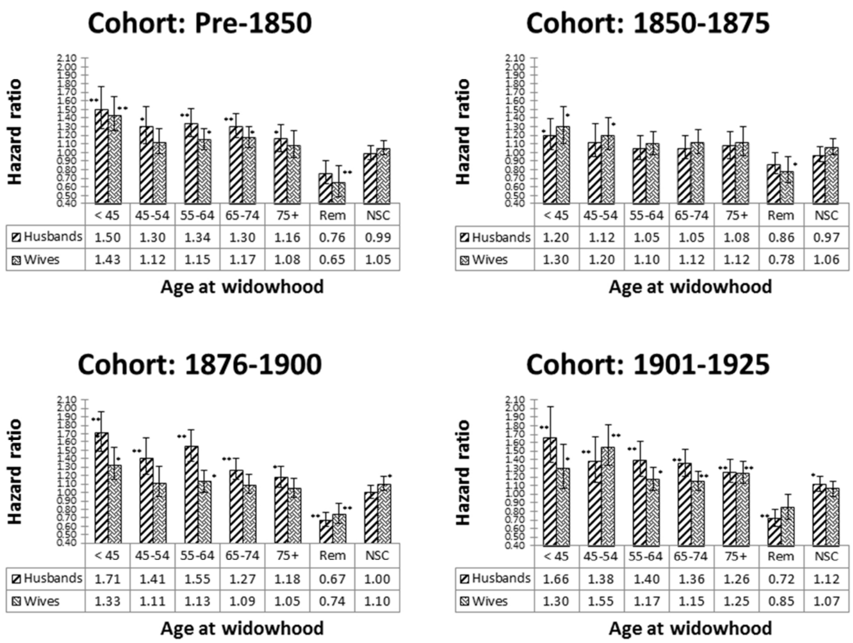

Figure 2. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).
173x233mm (300 x 300 DPI)

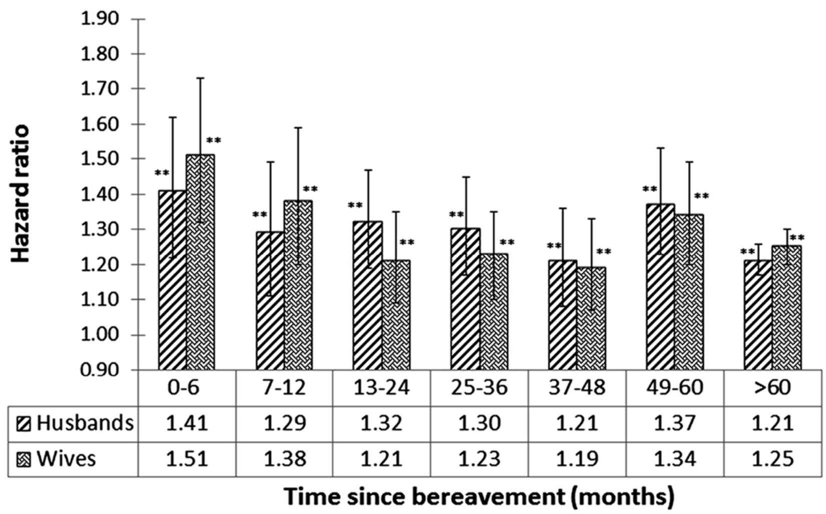

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).
173x233mm (300 x 300 DPI)

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

	Item No	Recommendation
Title and abstract	1	(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.
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found. See Abstract for findings.
Introduction		
Background/rationale	2	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.
Objectives	3	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.
Methods		
Study design	4	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]
Setting	5	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

		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	Summarise key results with reference to study objectives (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] 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.
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias (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.
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence (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). 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). 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.
Generalisability	21	Discuss the generalisability (external validity) of the study results (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. 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.

		Our study results indicate that remarriage plays an important role in improving the survival rate of widows and widowers in the Amish population.
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based (From Acknowledgments) This work was supported by research grants R01 HL69313, R01 DK54261, R01 AG1872801, R01 HL088119, R01 AR046838, and U01 HL72515 from the National Institutes of Health. This research was supported in part by the Intramural Research Program of the National Institutes of Health, NLM and the Geriatric Research and Education Center, Baltimore Veterans Administration Medical Center. These funders had no role in the study itself.

*Give information separately for exposed and unexposed groups.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at http://www.strobe-statement.org.

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: 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

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.

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.

- 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.

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 11]

The Amish maintain a cultural identity distinct from mainstream American

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

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. 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. The “individual table” of AGDB contains information about 539,822 individuals. The "relationship table” includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. 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. See Figure 1 for how the exclusion criteria were applied.

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. 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 analyses

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 divided into 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size.

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

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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.

- $ID1$: 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.

- $ID2$: 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.

- $ID3$: 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:

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

where $W_1,\ldots,W_5$ 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:

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

where $W_1,\ldots,W_7$ 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

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

Hazard Ratios

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

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 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.

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 bereavement and social support in the Amish population. 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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 3).[6 22] As expected, widowers 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.

Two strengths of our study are that, unlike many of the previously published studies,[5 13 16 20 21 24] we did not incorporate censoring methods into our analysis because of the high availability of death dates of the Amish widows and widowers in the first three cohorts; 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). Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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.

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

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

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

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.

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.

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

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Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups. Investigators can request access to AGDB by writing to AAS and to Dr. 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~~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

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.

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.

- 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.

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 11]

The Amish maintain a cultural identity distinct from mainstream American

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

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. The "individual table" of AGDB contains information about 539,822 individuals. The "relationship table" includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources

from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. 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. See Figure 1 for how the exclusion criteria were applied.

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. 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 analyses

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 ~~Hazards (CPH) model~~ 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 divided into 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size.

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

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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 (ID_1, ID_2 and ID_3) 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: $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, 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 $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, 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

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

Hazard Ratios

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

Figure ~~2~~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 ~~1~~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 ~~2~~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 ~~2~~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 ~~3~~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.

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). ~~Further, t~~The *deviance residual* plots revealed~~showed that there were~~ 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 bereavement and social support in the Amish population. 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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~~3).[6 22] As expected, widowers showed higher hazard ratios than widows except for the 1850-1875 cohort (Figure ~~1~~2).

In the present study, the association between bereavement and mortality is greater in the first 6 months for both men and women (Figure ~~3~~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 ~~3~~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.

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 we did not incorporate censoring methods into our analysis because of the high availability of death dates of the Amish widows and widowers in the first three cohorts; 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). Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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.

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

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. The lack of protective association was similarly observed when the number of surviving children was considered as a linear or as a categorical variable (data not shown). This contrasts with data from the Utah Population Database, in which increasing numbers of children were associated with a decreased hazard ratio.[8]

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

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

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.

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.

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

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Competing interests None.

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

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups.

Investigators can request access to AGDB by writing to AAS and to Dr. Biesecker.

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FIGURE LEGENDS

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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 12. 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

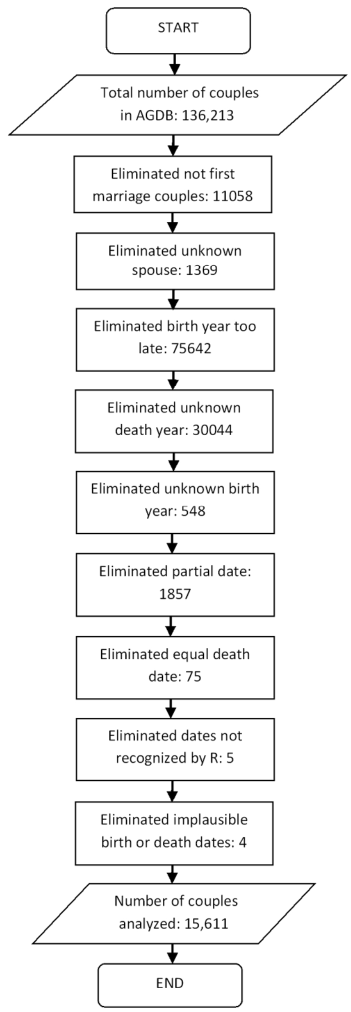
Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

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
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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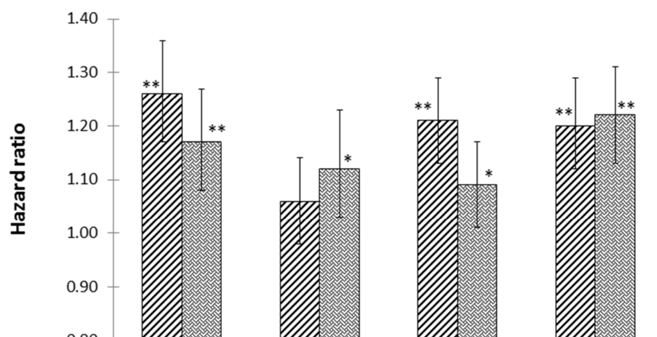

	Pre-1850	1850-1875	1876-1900	1901-1925
Husbands	1.26	1.06	1.21	1.20
Wives	1.17	1.12	1.09	1.22

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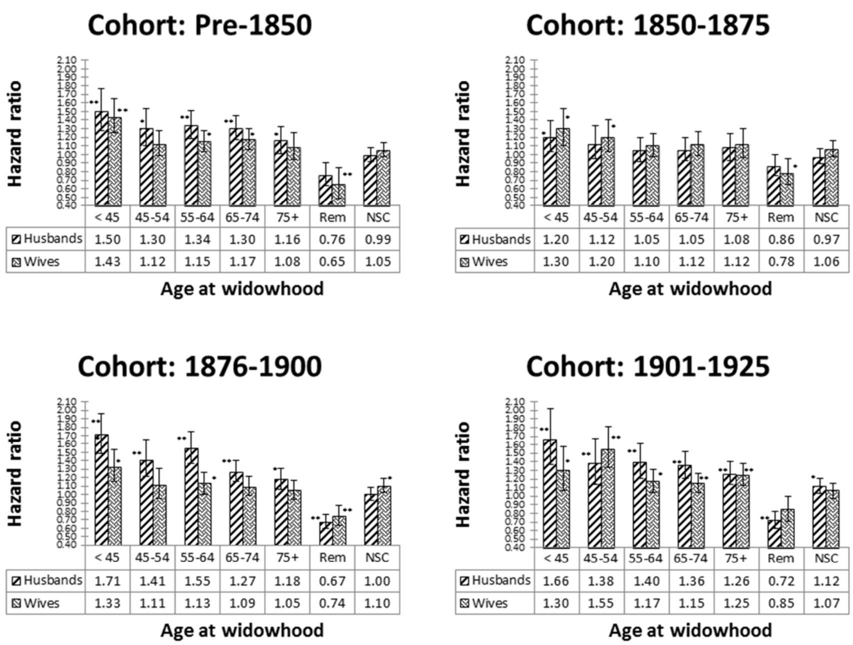

Figure 3. Hazard ratios of widowed husbands and wives versus their married counterparts according to age at widowhood. (design provided in Table 2b).
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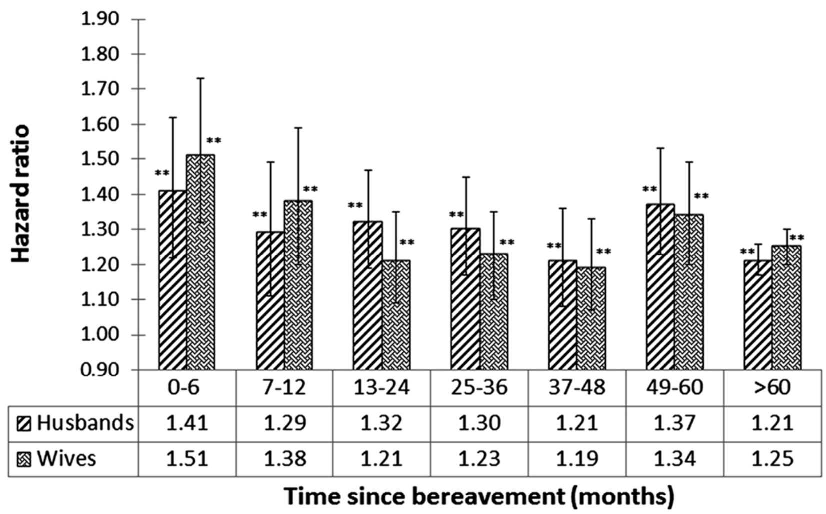

Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months). (design provided in Table 2c).
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Table S1: Wald statistics associated with models which are used to study the association 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	(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.
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found. See Abstract for findings.
Introduction		
Background/rationale	2	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.
Objectives	3	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.
Methods		
Study design	4	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]
Setting	5	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

		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	Summarise key results with reference to study objectives (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] 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.
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias (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.
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence (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). 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). 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.
Generalisability	21	Discuss the generalisability (external validity) of the study results (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. 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.

		Our study results indicate that remarriage plays an important role in improving the survival rate of widows and widowers in the Amish population.
Other information		
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based (From Acknowledgments) This work was supported by research grants R01 HL69313, R01 DK54261, R01 AG1872801, R01 HL088119, R01 AR046838, and U01 HL72515 from the National Institutes of Health. This research was supported in part by the Intramural Research Program of the National Institutes of Health, NLM and the Geriatric Research and Education Center, Baltimore Veterans Administration Medical Center. These funders had no role in the study itself.

*Give information separately for exposed and unexposed groups.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at http://www.strobe-statement.org.

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

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

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

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.

- 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 11]

The Amish maintain a cultural identity distinct from mainstream American

culture that is characterized by their traditional dress, a plain lifestyle, and non-adoption of modern technology (e.g., electricity, cars, telephones), German dialect, separate school system, and ultra-conservative Anabaptist religious practices. A central tenet of Amish culture throughout their history in the USA has been social cohesiveness with emphasis on family and community. Members of this tight-knit society have extraordinary social support from cradle to grave, including community-financing of medical costs and support during times of need. The community financing of medical costs became increasingly formalized during the 20th century to the point where it is now a formal system of self-insurance called "Amish Aid".

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and

further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. 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. The “individual table” of AGDB contains information about 539,822 individuals. The "relationship table” includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources

from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1901; (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 10,892 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were applied.

Our data set included 10,892 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood. For some analyses, these couples were first partitioned into three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and based on both husband's and wife's birth year (1876-1900) 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=291). Characteristics of these couples are shown in Table 1.

Statistical analyses

Similar to several past analyses,[2 8 13 16] we used the Cox Proportional Hazards (CPH) model where the response variable or survival time is 'age at 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. Husbands and wives 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size. To evaluate any possible bias by counting the number of surviving children at the death of the first spouse, we also estimated how many couples changed categories between the death of the first spouse and the death of second spouse due to death of one or more children during the widowhood period.

Survival analyses were performed using the R programming environment.[17] All estimates and confidence intervals were obtained using the `coxph` function

available in the `survival` package.

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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.

- $ID1$: 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.

- $ID2$: 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.

- $ID3$: 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: $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, and W_{75+}. Further, we also addressed the impact of remarriage and number of surviving children on the mortality of Amish individuals.

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

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

where $W_1,\ldots,W_5$ dummy variables are associated with $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, 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 $TSB_{0\text{-}6}$, $TSB_{7\text{-}12}$, … 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:

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

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

RESULTS

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

Hazard Ratios

The overall hazard ratios (HR), and their 95% confidence intervals (CIs), associated with widowhood are displayed in Figure 2 according to birth cohort. The significance of these hazard ratios are indicated with the help of p-values (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2. These hazard ratios were estimated using the CPH model and design provided in Table 2a. As expected, widowhood was associated with increased mortality for both husbands and wives following the death of spouse, with HRs ranging from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed husbands in cohort 1850-1875). The impact of widowhood was

disproportionately greater on surviving husbands than on surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little difference in mortality between widowed husbands vs. wives in the 1850-1875. There was no clear trend showing that the hazard ratios changed across cohorts in either surviving husbands or wives.

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.

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

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

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 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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 3).[6 22] As expected, widowers 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.

One strength of our study is that we could evaluate the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1) Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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 males and females across all cohorts . This association is likely influenced by the fact that males or females who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for males as compared to females, possibly reflecting support in Amish society for males getting remarried.

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

The implication of these findings is that differential familial support following bereavement does not appear to be the key factor affecting mortality increases in widowed Amish populations. One potential explanation is that spouses, as "attachment figures," provide a unique, irreplaceable social support which must be considered independently from other ancillary support providers, such as children.[24] Data suggesting that relative mortality risk is also elevated in divorced and never-married populations may support this hypothesis.[3] In addition, quality of social support has significantly greater effects on well-being than quantity of support in elderly populations of both sexes.[25]

In adults aged 65 and older, lower reported social support has been associated with decreased life satisfaction and increased depressive symptoms.[26] Family members and close friends should be especially vigilant during the sensitive acute period following the loss of a spouse, when relative mortality risk is the highest.

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

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

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Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups. Investigators can request access to AGDB by writing to AAS and to Dr. 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-value < 0.05 and p-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

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

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.

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

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.

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~~**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~~

~~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 socially cohesive Amish community. Remarriage is associated with a significant decrease in the mortality risk among Amish individuals.~~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.

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 11]

The Amish maintain a cultural identity distinct from mainstream American

culture that is characterized by their traditional dress, a plain lifestyle, and non-adoption of modern technology (e.g., electricity, cars, telephones), German dialect, separate school system, and ultra-conservative Anabaptist religious practices. A central tenet of Amish culture throughout their history in the USA has been social cohesiveness with emphasis on family and community. Members of this tight-knit society have extraordinary social support from cradle to grave, including community-financing of medical costs and support during times of need. The community financing of medical costs became increasingly formalized during the 20th century to the point where it is now a formal system of self-insurance called "Amish Aid".Members of this tight-knit society have extraordinary social support from cradle to grave, including community-managed health insurance and support during times of need. Elderly parents are taken care of by their children and neighbors; they do not use assisted living or nursing homes to care for their elderly.

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. 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. The “individual table” of AGDB contains information about 539,822 individuals. The "relationship table” includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1901; (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 10,892 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were applied.Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. 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. See Figure 1 for how the exclusion criteria were applied.

Our data set included 10,892 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood. For some analyses, these couples were first partitioned into three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and based on both husband's and wife's birth year (1876-1900) 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=291). Characteristics of these couples are shown in Table 1. 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. 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 analyses

Similar to several past analyses,[2 8 13 16] we used the Cox Proportional Hazards (CPH) model where the response variable or survival time is 'age at 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-

6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size. To evaluate any possible bias by counting the number of surviving children at the death of the first spouse, we also estimated how many couples changed categories between the death of the first spouse and the death of second spouse due to death of one or more children during the widowhood period. The number of surviving children was a time-independent covariate and divided into 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size.

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

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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 (ID_1, ID_2 and ID_3) 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: $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, and W_{75+}. Further, we also addressed the impact of remarriage and number of surviving children on the mortality of Amish individuals.~~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:

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

where $W_1,\ldots,W_5$ dummy variables are associated with $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, 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

We initially partitioned the 10,892 couples from AGDB into three cohorts ranging in size from 3,210 (husband birth year 1850-1875) to 3,971 (husband birth year 1876-1900). Wives were more likely to die after their husbands in all three cohorts while husbands had a higher mean age at death than their wives. The average age differences between the husbands and wives for all cohorts are shown in Table 1. The number of remarried wives was far smaller than the number of remarried husbands (n = 1,290 widowed husbands vs 378 widowed wives). The number and proportion of remarried husbands and wives were increased in the more contemporary cohorts (see Table 1). In contrast to other populations from the 19th and early 20th centuries,[4] the majority of widowed husbands did not remarry, making it interesting to study the effect of remarriage in the Amish population. 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 1876-1900), husbands had a higher mean age at death than their wives. The average age differences between the husbands and wives for all cohorts are shown in Table 1. The number of remarried wives was far smaller than the number of remarried husbands (n = 1,943 widowed husbands vs 613 widowed wives). The number and proportion of remarried husbands and wives were increased in the more contemporary cohorts (see Table 1). In contrast to other populations from the 19th and early 20th centuries,[4] the majority of widowed husbands did not remarry, making it interesting to study the effect of remarriage in the Amish population.

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Hazard Ratios

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

The significance of these hazard ratios are indicated with the help of p-values (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2. These hazard ratios were estimated using the CPH model and design provided in Table 2a. As expected, widowhood was associated with increased mortality for both husbands and wives following the death of spouse, with HRs ranging from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed husbands in cohort 1850-1875). The impact of widowhood was disproportionately greater on surviving husbands than on surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little difference in mortality between widowed husbands vs. wives in the 1850-1875. The impact of widowhood was disproportionately greater on surviving husbands than on surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little difference in mortality between widowed husbands vs wives in the 1850-1875 and 1901-1925 cohorts. There was no clear trend showing that the hazard ratios changed across cohorts in either surviving husbands or wives. Since no clear differences were observed between birth cohorts, all birth cohorts were combined to estimate the association between age at widowhood and mortality.

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). 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. 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 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).~~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. 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.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 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 bereavement and social support in the Amish population. 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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 3).[6 22] As expected, widowers 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.

One strength of our study is that we could evaluate the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1)~~Two strengths of our study are that, unlike many of the~~

previously published studies,[5 13 16 20 21 24] we did not incorporate censoring methods into our analysis because of the high availability of death dates of the Amish widows and widowers in the first three cohorts; 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). Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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 males and females across all cohorts . This association is likely influenced by the fact that males or females who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for males as compared to females, possibly reflecting support in Amish society for males getting remarried.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.

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

The implication of these findings is that differential familial support following bereavement does not appear to be the key factor affecting mortality increases in widowed Amish populations. One potential explanation is that spouses, as "attachment figures," provide a unique, irreplaceable social support which must be considered independently from other ancillary support providers, such as children.[2~~5~~4] Data suggesting that relative mortality risk is also elevated in divorced and never-married populations may support this hypothesis.[3] In addition, quality of social support has significantly greater effects on well-being than quantity of support in elderly populations of both sexes.[2~~6~~5]

In adults aged 65 and older, lower reported social support has been associated with decreased life satisfaction and increased depressive symptoms.[2~~7~~6] Family members and close friends should be especially vigilant during the sensitive acute period following the loss of a spouse, when relative mortality risk is the highest.

~~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.~~ 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.~~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.~~

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

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Competing interests None.

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Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups. Investigators can request access to AGDB by writing to AAS and to Dr. 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

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); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.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); NSC1: Number of Surviving Children (3-6 vs ≤ 2); NSC2: Number of Surviving Children (>6 vs ≤ 2); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-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).

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.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 10,892 spouse pairs according to birth cohort of husband.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)

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.

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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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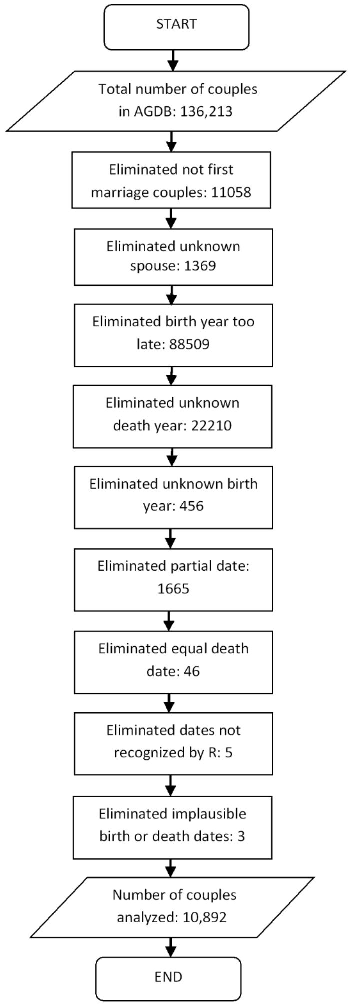

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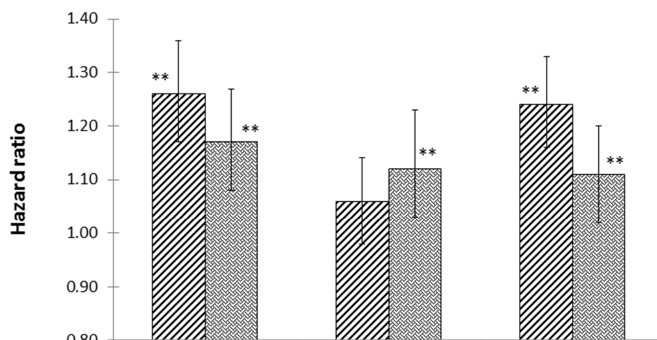

	Pre-1850	1850-1875	1876-1900
Husbands	1.26	1.06	1.24
Wives	1.17	1.12	1.11

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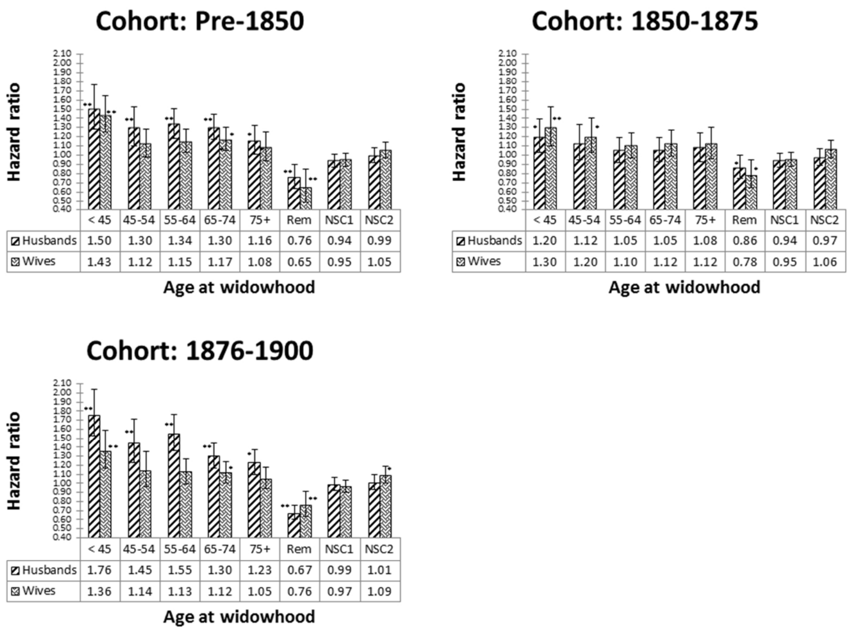

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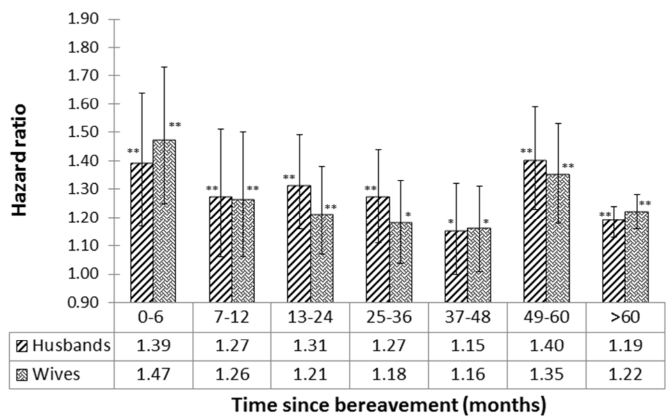

Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months) (design provided in Table 2c); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.
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Table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

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

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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

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

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.

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 11]

The Amish maintain a cultural identity distinct from mainstream American

culture that is characterized by their traditional dress, a plain lifestyle, and non-adoption of modern technology (e.g., electricity, cars, telephones), German dialect, separate school system, and ultra-conservative Anabaptist religious practices. A central tenet of Amish culture throughout their history in the USA has been social cohesiveness with emphasis on family and community. Members of this tight-knit society have extraordinary social support from cradle to grave, including community-financing of medical costs and support during times of need. The community financing of medical costs became increasingly formalized during the 20th century to the point where it is now a formal system of self-insurance called "Amish Aid".

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and

further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. 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. The "individual table" of AGDB contains information about 539,822 individuals. The "relationship table" includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources

from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1901; (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 10,892 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were applied.

Our data set included 10,892 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood. For some analyses, these couples were first partitioned into three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and based on both husband’s and wife’s birth year (1876-1900) 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=291). Characteristics of these couples are shown in Table 1.

Statistical analyses

Similar to several past analyses,[2 8 13 16] we used the Cox Proportional Hazards (CPH) model where the response variable or survival time is ‘age at 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. Husbands and wives 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size. To evaluate any possible bias by counting the number of surviving children at the death of the first spouse, we also estimated how many couples changed categories between the death of the first spouse and the death of second spouse due to death of one or more children during the widowhood period.

Survival analyses were performed using the R programming environment.[17] All estimates and confidence intervals were obtained using the `coxph` function

available in the `survival` package.

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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 (ID_1, ID_2 and ID_3) 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: $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, and W_{75+}. Further, we also addressed the impact of remarriage and number of surviving children on the mortality of Amish 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 $W_{<45}$, $W_{45\text{-}54}$, $W_{55\text{-}64}$, $W_{65\text{-}74}$, 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 $TSB_{0\text{-}6}$, $TSB_{7\text{-}12}$, … 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 $TSB_{0\text{-}6}$, $TSB_{7\text{-}12}$, $TSB_{13\text{-}24}$, $TSB_{25\text{-}36}$, $TSB_{37\text{-}48}$, $TSB_{49\text{-}60}$ and $TSB_{>60}$ columns provided in the Table 2c.

RESULTS

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

Hazard Ratios

The overall hazard ratios (HR), and their 95% confidence intervals (CIs), associated with widowhood are displayed in Figure 2 according to birth cohort. The significance of these hazard ratios are indicated with the help of p-values (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2. These hazard ratios were estimated using the CPH model and design provided in Table 2a. As expected, widowhood was associated with increased mortality for both husbands and wives following the death of spouse, with HRs ranging from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed husbands in cohort 1850-1875). The impact of widowhood was

disproportionately greater on surviving husbands than on surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little difference in mortality between widowed husbands vs. wives in the 1850-1875. There was no clear trend showing that the hazard ratios changed across cohorts in either surviving husbands or wives.

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.

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

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 bereavement and social support in the Amish population. 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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 3).[6 22] As expected, widowers 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,[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 that is unmasked in the absence of spousal support.

One strength of our study is that we could evaluate the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1) Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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 males and females across all cohorts . This association is likely influenced by the fact that males or females who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for males as compared to females, possibly reflecting support in Amish society for males getting remarried.

Interestingly, more children at the time of the death of the first spouse was associated with increased the risk of death, though the hazard ratio for having > 6 surviving children as compared to ≤ 2 was not significantly greater than 1.0. This result does not support the hypothesis that more surviving children confer a survival advantage to parental longevity, as perhaps by providing social support for their parents. Spouses in the Amish society may also provide unique emotional, psychological, and social support to each other which cannot be provided by their surviving children. The lack of protective association was similarly observed when the number of surviving children was considered as a linear or as a categorical variable (data not shown). This contrasts with data from the Utah Population Database, in which increasing numbers of children were associated with a decreased hazard ratio.[8] We considered the number of surviving children as a separate term (Table 2a), but did not evaluate the interaction of number of surviving children with widowhood.

The implication of these findings is that differential familial support following bereavement does not appear to be the key factor affecting mortality increases in widowed Amish populations. One potential explanation is that spouses, as "attachment figures," provide a unique, irreplaceable social support which must be considered independently from other ancillary support providers, such as children.[24] Data suggesting that relative mortality risk is also elevated in divorced and never-married populations may support this hypothesis.[3] In addition, quality of social support has significantly greater effects on well-being than quantity of support in elderly populations of both sexes.[25]

In adults aged 65 and older, lower reported social support has been associated with decreased life satisfaction and increased depressive symptoms.[26] Family members and close friends should be especially vigilant during the sensitive acute period following the loss of a spouse, when relative mortality risk is the highest.

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

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Competing interests None.

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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-value < 0.05 and p-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

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

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.

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

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

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

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 11]

The Amish maintain a cultural identity distinct from mainstream American

culture that is characterized by their traditional dress, a plain lifestyle, and non-adoption of modern technology (e.g., electricity, cars, telephones), German dialect, separate school system, and ultra-conservative Anabaptist religious practices. A central tenet of Amish culture throughout their history in the USA has been social cohesiveness with emphasis on family and community. Members of this tight-knit society have extraordinary social support from cradle to grave, including community-financing of medical costs and support during times of need. The community financing of medical costs became increasingly formalized during the 20th century to the point where it is now a formal system of self-insurance called "Amish Aid".

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. Previous studies used covariates such as education (homogeneous in the Amish),[12] health habits, remarriage, church visits, neighborhood interaction, and household size to study the relationship between bereavement and social support.[4 8 13]

We estimated bereavement effects in widows and widowers separately, and

further assessed mortality risks as a function of age at widowhood and time since bereavement.[2 8] 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.

MATERIAL AND METHODS

Data source

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. 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. The “individual table” of AGDB contains information about 539,822 individuals. The "relationship table” includes information about 136,213 Amish couples. An individual who is married multiple times participates in multiple relationship table entries. There are 1,369 relationship entries among the 136,213 entries concerning children for whom one or both biological parents are unknown (Figure 1).

Amish people rarely give birth out of wedlock, and a vast majority of the mating samples we used are documented as marriages in the three original sources

from which AGDB is derived. However, there is less availability of marriage dates than birth dates; we did not wish to exclude couples with missing marriage dates. For this study, we included couples in which both spouses met four conditions: (1) they were born prior to January 1, 1901; (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 10,892 couples met the inclusion criteria. The oldest person included was born on February 14, 1725, and the youngest on December 30, 1900. See Figure 1 for how the exclusion criteria were applied.

Our data set included 10,892 couples with information on their date of birth, date of death, number of surviving children, family IDs, and remarriage status after widowhood. For some analyses, these couples were first partitioned into three cohorts based on the husband's birth year (prior to 1850 and 1850-1875) and based on both husband's and wife's birth year (1876-1900) 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=291). Characteristics of these couples are shown in Table 1.

Statistical analyses

Similar to several past analyses,[2 8 13 16] we used the Cox Proportional Hazards (CPH) model where the response variable or survival time is 'age at 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. Husbands and wives 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, 3-6 children, and > 6 children are separate. These boundaries were chosen to ensure categories that were roughly balanced in size. To evaluate any possible bias by counting the number of surviving children at the death of the first spouse, we also estimated how many couples changed categories between the death of the first spouse and the death of second spouse due to death of one or more children during the widowhood period.

Survival analyses were performed using the R programming environment.[17] All estimates and confidence intervals were obtained using the `coxph` function

available in the `survival` package.

Representation of survival data for CPH analysis

We show below examples of how we represented the survival data for CPH analysis using a time modeling approach. To represent the survival data columns, the 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.

- $ID1$: 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.

- $ID2$: 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.

- $ID3$: 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 the mortality of Amish individuals.

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

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

where $W_1,\ldots,W_5$ 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:

$h(t) = h_0(t) exp[\sum \beta_j W_j]$ with j = 1,…,7

where $W_1,\ldots,W_7$ 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

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

Hazard Ratios

The overall hazard ratios (HR), and their 95% confidence intervals (CIs), associated with widowhood are displayed in Figure 2 according to birth cohort. The significance of these hazard ratios are indicated with the help of p-values (*:p-value<0.05 and **:p-value<0.001) on the top of each block in Figure 2. These hazard ratios were estimated using the CPH model and design provided in Table 2a. As expected, widowhood was associated with increased mortality for both husbands and wives following the death of spouse, with HRs ranging from 1.06 to 1.26 (all HRs significantly greater than 1 except for widowed husbands in cohort 1850-1875). The impact of widowhood was

disproportionately greater on surviving husbands than on surviving wives in the pre-1850 and 1876-1900 cohorts, although there was little difference in mortality between widowed husbands vs. wives in the 1850-1875. There was no clear trend showing that the hazard ratios changed across cohorts in either surviving husbands or wives.

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.

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

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 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 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]

Several previous studies on American, European, and Middle Eastern populations have found that mortality is magnified in individuals widowed at a younger age and that widowers have higher mortality risk than widows.[3 5 8 16 20 22] The LDS study is closest to our study because of the large family sizes and the population selected as a religious isolate.[8] 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 3).[6 22] As expected, widowers 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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that is unmasked in the absence of spousal support.

One strength of our study is that we could evaluate the effect of remarriage because there were substantial numbers of widowed individuals who did and did not get remarried (Table 1) Shor et al. noted that the difficulty in studying the effect of remarriage in populations where all individuals are deceased because "In previous decades, widowed men almost always remarried".[4] 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 males and females across all cohorts . This association is likely influenced by the fact that males or females who get remarried are in sufficiently good physical and mental health to do so. Further, the survival rate is higher for males as compared to females, possibly reflecting support in Amish society for males getting remarried.

Interestingly, more children at the time of the death of the first spouse was associated with increased the risk of death, though the hazard ratio for having > 6 surviving children as compared to ≤ 2 was not significantly greater than 1.0. This result does not support the hypothesis that more surviving children confer a survival advantage to parental longevity, as perhaps by providing social support for their parents. Spouses in the Amish society may also provide unique emotional, psychological, and social support to each other which cannot be provided by their surviving children. The lack of protective association was similarly observed when the number of surviving children was considered as a linear or as a categorical variable (data not shown). This contrasts with data from the Utah Population Database, in which increasing numbers of children were associated with a decreased hazard ratio.[8] We considered the number of surviving children as a separate term (Table 2a), but did not evaluate the interaction of number of surviving children with widowhood.Interestingly, increasing numbers of surviving children at the time of widowhood did not confer a survival advantage for Amish individuals. This result was counter to our hypothesis that children can help provide social support for their parents. The hazard ratio was greater than 1.0 (but not significant) for all Amish individuals with number of surviving children > 6 as compared to ≤ 2. Spouses in the Amish society may also provide unique emotional, psychological, and social support to each other which cannot be provided by their surviving children. The lack of protective association was similarly observed when the number of surviving children was considered as a linear or as a categorical variable (data not shown). This contrasts with data from the Utah Population Database, in which increasing numbers of children were associated with a decreased hazard ratio.[8]

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The implication of these findings is that differential familial support following bereavement does not appear to be the key factor affecting mortality increases in widowed Amish populations. One potential explanation is that spouses, as "attachment figures," provide a unique, irreplaceable social support which must be considered independently from other ancillary support providers, such as children.[24] Data suggesting that relative mortality risk is also elevated in divorced and never-married populations may support this hypothesis.[3] In addition, quality of social support has significantly greater effects on well-being than quantity of support in elderly populations of both sexes.[25]

In adults aged 65 and older, lower reported social support has been associated with decreased life satisfaction and increased depressive symptoms.[26] Family members and close friends should be especially vigilant during the sensitive acute period following the loss of a spouse, when relative mortality risk is the highest.

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

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Competing interests None.

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Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement Data from the AGDB are available to investigators (including ARS and various others not participating in this study) who have an IRB-approved protocol to study the Amish or other Anabaptist groups. Investigators can request access to AGDB by writing to AAS and to Dr. 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-value < 0.05 and p-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

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

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.

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

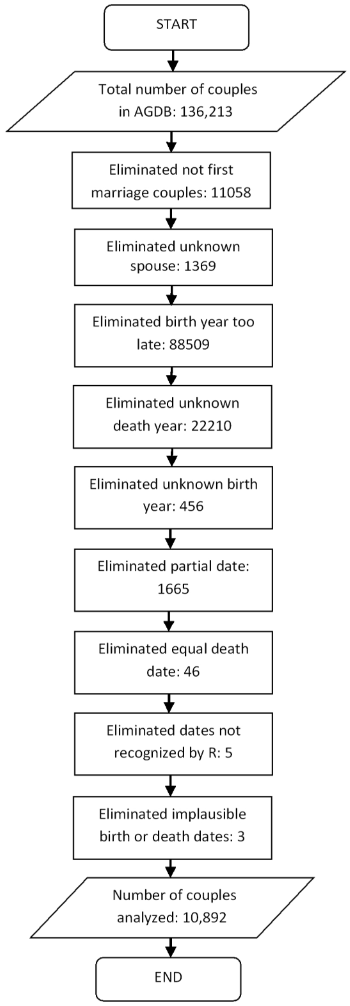
Table 2b: Data structure for Cox Proportional Hazard model that estimates association between widowhood at given ages and mortality.

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



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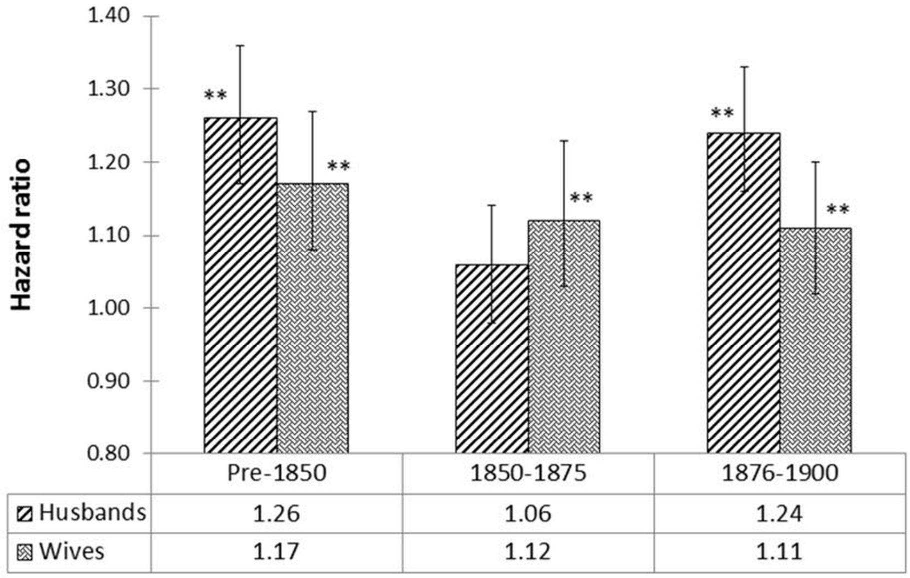

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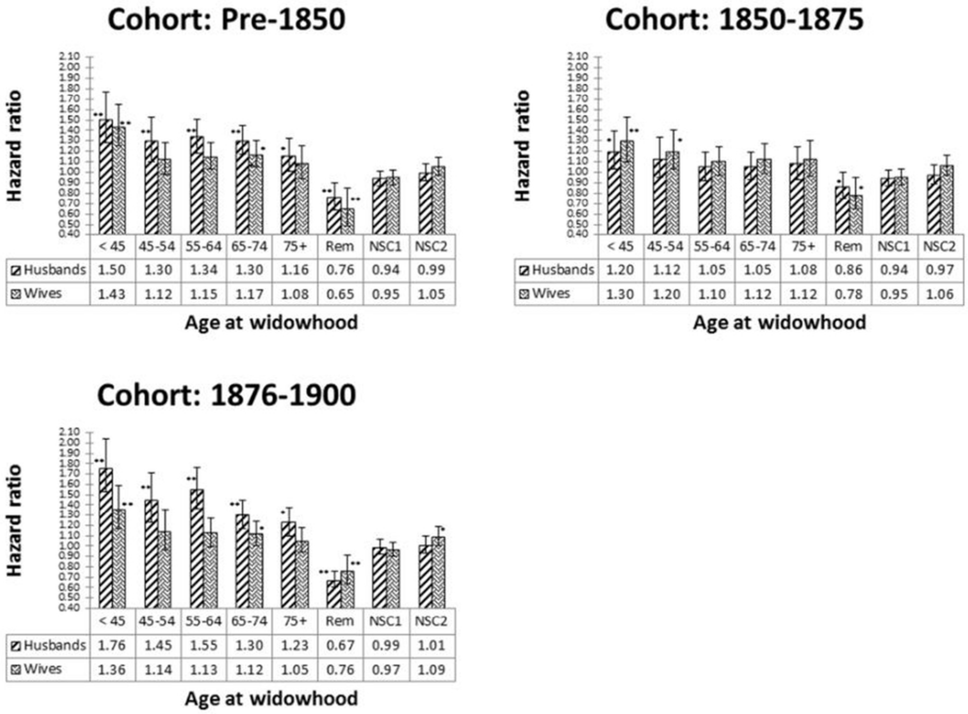

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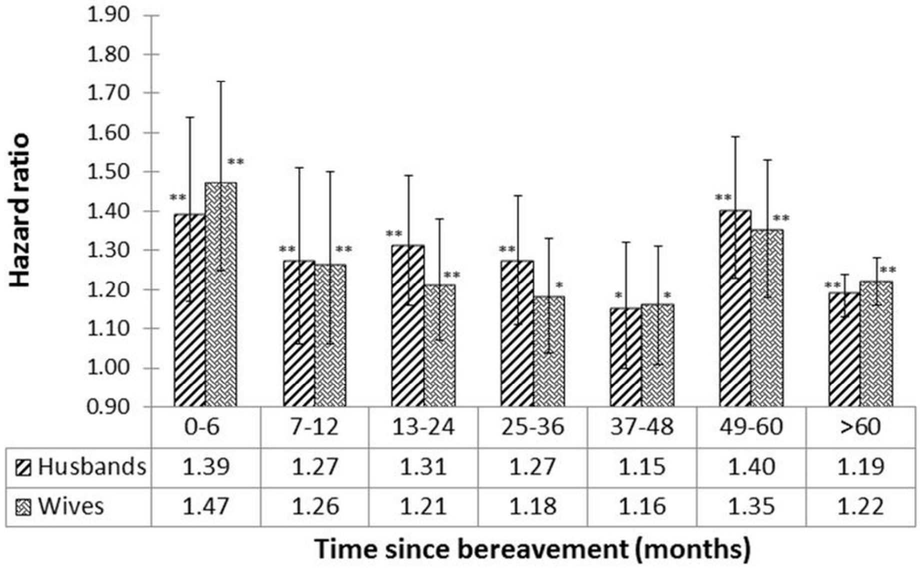

Figure 4. Hazard ratios of widowed husbands and wives versus their married counterparts according to time since bereavement (months) (design provided in Table 2c); * and ** on the top of the blocks represent significance of hazard ratios with p-value < 0.05 and p-value < 0.001, respectively.
149x90mm (300 x 300 DPI)

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	