LONGEVITY: A FAMILY MATTER? INSIGHTS FROM AN INLAND VILLAGE OF SARDINIA (ITALY), 1850–2010[1]

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Abstract. Familial transmission of longevity is a primarily studied topic in human longevity research. The contribution of the genetic component inherited as part of the shared family DNA is hard to disentangle from that of the familiar environment. Based on the analysis of a genealogical database of an in-land village in Sardinia, this study aims to provide insights into the possible role of familial transmission of survival, estimating the effects of parental loss and parental longevity in two significant moments of individual life: in early childhood and at older ages. Using event history methods, survival trajectories of individuals born during the period 1850-1910 in a village of Sardinia (Italy) were analysed. It emerged that effects of parents' survival occur both in the short and long term. Mother's death before five years of age increases the mortality rates of the offspring, which are mitigated by the presence of at least one sister who can replace her in the care tasks. At older ages, only maternal survival is significant for Ego's longevity. The findings point to the relevance of the genetic component. Still, it suggests reflecting on the importance of the social dimension, the possible role of care and the transmission of knowledge and cultural capital, and social networks.

1. Introduction

Studies focusing on the relationship between familial transmission and individual survival are innumerable, all to understand the mechanisms by which genetic heritage transmitted from parents to children acts on survival, both concerning biological characteristics, the possible relationship with diseases, and longevity (see among others, Kerber *et al.*, 2001; Perls *et al.* 2007; You *et al.* 2010; O'Brein *et al.*, 2018; van den Berg *et al.* 2017; 2018). However, members of the same family also share the family environment, namely specific household characteristics, daily

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habits, diet, the physical environment and climate, everyday experiences, and so on (Salaris, 2010; Jarry *et al.*, 2012; Iachine *et al.*, 1998). Genetic and environmental factors often overlap and are difficult to operationalize, and researchers frequently work with incomplete data.

Family often represents a privileged area of investigation for human survival studies as its members share pieces of their genetic heritage and environment. Its traits significantly affect health status and survival, even at older ages. Relevant characteristics include family size and structure, birth order, the presence or absence of parents at critical periods, living arrangements, wealth, and so on (van Poppel and Liefbroer, 2005). Characteristics of families of origin may persistently affect survival, as events at different life stages – even early ones – impact people's health across lifetimes through an accumulation process or as a long-term consequence of compromising early life damage (Ben-Shlomo and Kuh, 2002).

Studies on the parent-child relationship represent the most consistent body of research, pointing out that the parental effect on offspring survival is complex, and estimates might be affected by the adopted longevity threshold. Cournil *et al.* (2000) for example, reported a positive association between the survival of fathers and that of their offspring, with a marked effect father-daughter at advanced ages. Studies conducted on different population instead reported no statistically significant effect of paternal survival (Mazan and Gagnon, 2007). While, in Sardinia, a more significant maternal effect has been observed (Caselli *et al.*, 2006), pointing for the possible influence from genes and maternally inherited mitochondrial DNA.

This study aims to provide insights into the possible mechanism of familial transmission of survival, trying to estimate the effects of parental loss and parental survival in two significant moments of individual life, namely in early childhood and at older ages, in a bid to answer the following questions:

- In childhood, does the loss of the father affect the survival of offspring? And what is the effect of the loss of the mother?
- In the first five years of life, are the possible adverse effects of the mother's loss mitigated by the presence of other female figures within the family?
- Is there any survival resemblance between parents and offspring? Are long-lived individuals' children of long-lived parents? And at older ages, does the presence of other female figures significantly affect survival?

This work used data derived from the Villagrande Longevity Database (VILD), focusing on all the children (2,173 individuals) born in the municipality of Villagrande Strisaili in Sardinia (Italy) from the 444 marriages celebrated during 1850-1910 for which survival until 2010 is known. Villagrande Strisaili is renowned for the exceptional longevity of its population (Poulain *et al.*, 2004; 2011; Salaris *et al.*, 2013; Salaris, 2015).

The first part of this paper presents a review of studies and related theoretical references on familial transmission of longevity. While the second part describes the data source and quality and the applied methods, followed by the survival analysis results. Finally, the discussion section highlights relevant insights that could contribute to the general debate on the familiar transmission of individual survival, which is genetic but also influenced by social dimension.

2. Materials and methods

2.1. Data sources, data quality, and population under study

Data from the analysis derives from Villagrande Longevity Database (VILD), a three-generation level genealogical database of the municipality of Villagrande Strisaili in Sardinia (Italy), which includes 7,250 individuals, where data was validated to reduce biases deriving from under-registration of births, deaths and migration, age misreporting, and selective censoring (for more details, see Salaris, 2010). The target population of this work initially included 2,306 individuals born from the marriages celebrated from 1850 to 1910, considering only the unions within which there have been births (444 out of 499 marriages). The analysis was restricted to individuals for which survival was known until 2010. Accordingly, 2,173 cases were finally considered for analysis (94.2% of total newborns). Survival resemblance between parents and offspring is investigated, focusing on post-reproduction survival of 50-year-old individuals (1,043 cases). It's worth noting that individuals under study are not interdependent as Ego belonging to birth cohorts from 1850 to 1890 are in large measure included in the analysis as parents of Ego born in the next generation.

2.2. Familial resemblance, longevity threshold and variables

To identify groups of survivors for fathers and mothers of Ego, longevity thresholds were estimated using a Quantile Regression Model (QRM) (Koenker and Bassett, 1978). Table 1 reports the QRM estimates. The median age of Ego is 40.39 for males and 51.16 for females. Gender differences can be attributed to the selective effects of male mortality related to war and accidental death (Salaris, 2015).

Ego age at death, Ego parent age at death, and Ego age at the time of loss of his or her mother or father are all continuous in the extended Cox proportional hazard model, which according to the code assigned to each family, includes family-shared frailty (Therneau and Grambsch, 2000). Models' estimates were controlled for birth

cohort effects as high survival level were observed for individuals born between 1902 and 1911 (Salaris, 2014).

Table 1 – *Age at death of Ego, mothers, and fathers according to specific quantiles*

Quantiles	Age at death of Ego Males	Age at death of Ego Female	Maternal age at death	Paternal Age at death
0.10	0.76	0.95	38.17	48.65
0.20	2.37	2.94	48.15	58.70
0.50	40.39	51.16	71.40	69.91
0.75	78.90	82.08	80.59	80.01
0.80	82.73	84.20	82.13	82.91
0.90	89.76	89.24	87.14	87.70

2.3. Methods

Firstly, infant mortality rates were estimated. Subsequently, the non-parametric approach of Kaplan–Meier (KM) survival estimation (Kleinbaum, 1995) compared selected groups of survivors. Differences within categories were tested for significance using Breslow and Log Rank tests (Blossfeld and Rohwer, 2002). A series of extended Cox proportional hazard models were fit. They included frailty and the birth cohort as control variables. The introduction of shared frailty allows to account for unobserved factors affecting individual risk of mortality, which are assumed to be equally shared with others in the family (Wienke, 2003; Garibotti *et al.*, 2006). Shared frailty is assumed to be gamma-distributed (Therneau and Grambsch, 2000). All covariates met the proportionality assumption, tested using Schoenfeld residuals (Schoenfeld, 1982).

3. Results

3.1. Infancy and mortality. Is survival an early family issue?

Table 2 reports the mortality rates (mx) estimates for all newborns in Villagrande Strisaili (1850-1910). Estimates distinguish between the 'survival category' of parents, namely whether the Ego mother and father belonged to the first 20 percentile of parent survivors ('frail') or the longest-living ones (≥80 percentile, 'robust'). The estimates suggest that the effect of a father's survival on children is limited,

regardless if he is 'frail' or 'robust'. Children of frail mothers have higher mortality rates, which at birth are at 183.0 per thousand (+66%) against an average value of 112.7. These estimates, especially during the first year of life, should be considered with caution as they include cases of maternal mortality and more generally, the available data do not allow to discern among the possible influences of environmental conditions and associated diseases.

Between the first and second birthday, m_x equals 126.9 per thousand (+70%) against an average of 74.4 observed for all infants born in the village. However, this disadvantage attenuates and disappears at subsequent ages. Still, despite some differences, when considering the most robust cluster of mothers, their children prove to be advantaged among their peers.

Table 2 – *Mortality rates (m_x) of all Villagrande Strisaili newborns from birth to age five by parental survival group (value ‰)*

Age	ALL	Std. Er	FRAIL (20%)		ROBUST (20%)	
			Mother	Father	Mother	Father
0	112.7	7.4	183.0	122.6	110.1	116.3
1	74.4	6.3	126.9	63.4	29.9	71.2
2	35.4	4.5	46.2	58.5	33.8	24.0
3	23.5	3.7	23.9	16.0	11.5	24.5
4	18.0	3.3	21.0	29.5	17.5	15.6

Kaplan–Meier (KM) curves help to understand the evolution of mortality trajectories and how the loss of a parent acts on survival estimates at all ages. Figures 1a and 1b clearly show that the loss of one of the parents might be considered a stressful event with a negative impact on individual survival. Effects are more marked from birth to early adulthood but persist at all ages. The mother's loss appears to have a greater impact than that of the father.

And what could be the role played by the presence of other female figures when Ego loses their mother? Figures 2a and 2b compare cumulative survival curves of individuals who had (or not) at least one older sister (aged ten years and over) or one aunt (in the maternal kinship line) when they lost their mothers before their fifth birthdays. Differences between curves in Figure 2b are not statistically significant. In the case of the presence of an aunt, several crossover points occur, which casts doubt about the positive effect of these female figures' presence on Ego's survival. However, the presence of an older sister proves to have a protective effect on Ego survival (Figure 2a).

Figure 1 – *Kaplain-Meier survival curve of Villagrande Strisaili new-borns having or not lost before five years of age*

a) their mother *b) their father*

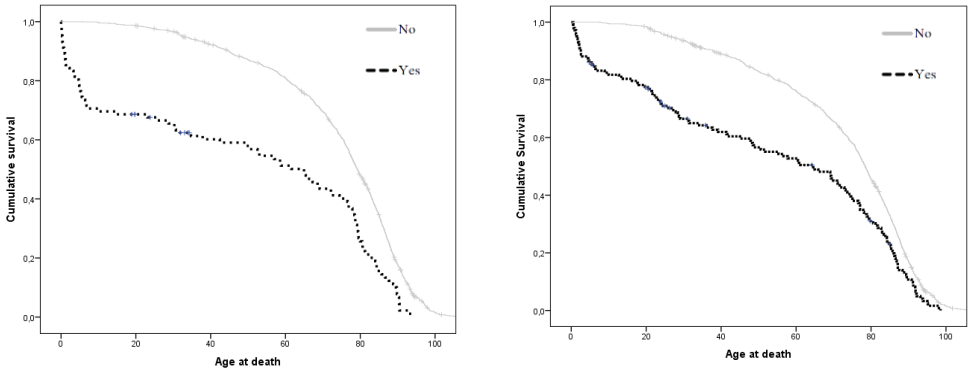
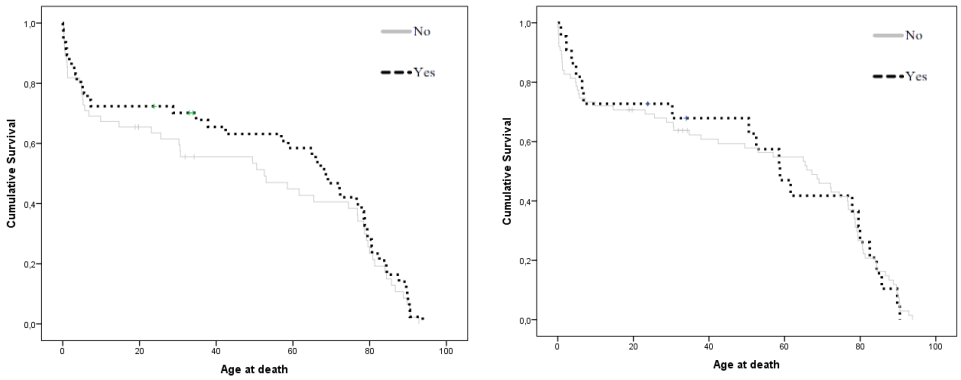


Figure 2 – *Kaplain-Meier survival curve of Villagrande Strisaili new-borns having or not at their mother's death occurring before five years of age*

a) at least one older sister *b) at least one aunt*



3.2. Effect of familial transmission of survival at post-reproduction ages

Focusing now on the possible effects of parental survival on the post-reproductive survival of their offspring, a set of extended Cox proportional hazard models were

fit. Estimates are presented in Table 3. All models have an Ego birth cohort as a control variable, and family-shared frailty frames individuals into their family of origin. Preliminary Model 1 estimates the impact of parental age at death on all newborns included in this study (2,173 cases). Estimates showed no sex effects and maternal and paternal age at death proved insignificant for Ego survival during the entire life span. Only losing the mother before age five proves to have a reducing effect on Ego's probability of survival, meaning that the adverse effects of the mother's death before five years, already detected when analyzing early survival, persist during the Ego lifetime.

Table 3 – *Extended Cox proportional hazard odds ratio of survival for Villagrande Strisaili from birth and age 50+.*

COVARIATES	Model 1 Basic	Model 2 Basic	Model 3 Adding females figures
	ALL newborns	50+ newborns	50+ newborns
Sex			
Males (ref.)	1	1	1
Females	1.012	1.013	1.070
Age at death of father (continuous)	0.989	1.008	0.990
Age at death of mother (continuous)	1.006	1.000	1.038
Age of Ego at loss of father (continuous)	1.012	1.012	1.040
Age of Ego at loss of mother (continuous)	0.982*	1.008	0.987**
Having at least 1 older sister			
yes			1
no			0.895
Having at least 1 aunt			
yes			1
no			0.914

Legend: *p<0.05; **p<0.01; ***p<0.001

More importantly, the focus shifts to mortality trajectories in post-reproductive ages (1,043 cases). As Model 2 shows, none of the variables included in the model prove to be statistically significant. Separate models were run for males and females, which didn't detect any statistically significant effect of parental death on their

offspring. For the sake of space, the results are not reported here. Although the number of cases is quite limited, Model 3 further explains the possible role of familial characteristics in this environmental dimension. The presence of at least one older sister or aunt when Ego lost their mother before five years of age was introduced into the model. Estimates show that the timing of the mother's loss becomes statistically significant for Ego once the model considers the role of other female family members. This finding cautiously suggests that the presence of an older sister might be viewed as a person of support when a traumatic event, such as the loss of the mother, occurs early in Ego's life.

4. Discussion

This work aimed to investigate the possible effects of the ages at death of parents on their offspring's survival at different stages of their lives, namely early childhood and later life after age 50. Survival trajectories analysis of individuals born in Villagrande Strisaili suggests that between the survival of the parents and that of the offspring, there is an observable link at all ages. However, the maternal bond is the one for which the greatest and, in some cases, statistically significant effects are observed, with no gender effect, confirming prior finding (Salaris, 2010; Salaris *et al.*, 2013).

In the examined community, a traumatic event such as the mother's death before age five for Ego proves to have a relevant impact. This finding makes us think that biological vulnerability, genetic factors, and common pathologies between mother and child lead to higher-than-average mortality rates. Estimates of infant mortality show that the children of frail mothers record higher mortality rates, which at birth are at 183.0 per thousand (+66%) against an average value of 112.7. Deaths between the first and second birthdays are 126.9 per thousand (+70%) versus the average of 74.4 observed for all infants born in the village.

However, higher mortality could also be related to a lack of care when the mother dies. She is the one primarily entrusted with caring for the child. Who takes care of the baby when the mother dies? Support in this direction can be traced in the findings, albeit cautiously, given the reduced number of cases. It derived from analyzing the possible effect on infant mortality estimates of so-called 'alternative' female figures such as one sister older than Ego or at least one aunt (on the maternal line). The presence of one sister in Villagrande Strisaili families - when mother prematurely died - mitigated the adverse effect that such a traumatic event inevitably brought with it.

The parents' survival effect on their children has also been observed in post-reproduction survival in the population studied here. The relationship with the

mother's survival registers the only statistically significant results in the estimates of the Cox proportional hazard models. These findings somehow confirm the indications derived from previous studies and in other populations on the importance of the maternal role for the Ego's survival. However, how could this result be interpreted in the context of Villagrande Strisaili? Providing a genetic explanation can be the most direct choice. But still, in our conclusive reflections, we would like to draw attention to environmental components and the possible role of cultural capital focusing on this select maternal line of transmission. The possible validity of this explanation lies in the fact that most frequent models of cultural transmission are also vertical as genetic ones. Parents transmit to children what they know, from having learned it from their parents, adding what they have discovered, invented, or learned during their existence. Transmission models of knowledge are difficult to distinguish from genetic ones, as they both occur among the same actors and involve a certain resemblance between parents and children or relatives in general (Cavalli Sforza, 2004). Hence, the ability to survive could be part of individual cultural capital, namely the accumulation of knowledge, behaviors, and skills which constitute a resource from which individuals build their cultural competence and social standing. Through contact and interaction, parents transmit their knowledge, allowing their children to learn. Cultural capital exists in an embodied state, namely, the individual identified by the self (Bourdieu, 1986). It is consciously acquired over the life course and passively assimilated through education and socialization of culture and traditions. Offspring survival is time; in our case, the time spent with mothers could also be interpreted as exposure time for acquiring knowledge and valuable skills for survival. The longer the survival of mothers, the longer the available time for children to learn, practice, and assimilate traditions, remedies, and food preparation, which is favorable for their survival. Under this possible interpretation, findings suggest that in Sardinia, the presence of a mother assumes a key role in the survival of the offspring, which might be related not exclusively to genetic endowment. They also transmit to their offspring cultural capital, which is enriched by the role of social networks that are prerogatives of female figures. The relevance of this aspect, in Villagrande Strisaili, can be hypothesized to be accentuated by the cyclical absence of the fathers' transhumant shepherds, which gives mothers a central role in the lives of their offspring. More generally, aware that distinguishing between genetic and environmental effects is not clear-cut, we believe that the latter cannot be discounted as factors of secondary importance. Instead, the observed differential mortality in the Villagrande Strisaili population estimates invites a reflection on the transmission of cultural capital (knowledge and skills) and social networks that would require further investigation.

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