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A POPULATION WHERE MEN LIVE AS LONG AS WOMEN:

VILLAGRANDE STRISAILI (SARDINIA)

Michel POULAIN¹, Gianni PES², Luisa SALARIS³,

¹ GÉDAP – Université Catholique de Louvain - Belgium

² Department of Biomedical Sciences – Università degli Studi di Sassari - Italy

³ GÉDAP – Université Catholique de Louvain – Belgium ; Università degli Studi di Cagliari - Italy

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Michel POULAIN⁴, Gianni PES⁵, Luisa SALARIS⁶,

Corresponding author: Michel Poulain

E-mail: michel.poulain@uclouvain.be

Introduction

In developed countries, it has been widely documented that females live longer than males (Hazzard 1994, Crose 1997, Clarke 2000). This so called “female advantage” in survival means that throughout the lifespan, female death rates are usually lower than those recorded for males (Daw 1961, Philips 2005).

In traditional societies, differences in mortality rates between males and females were less pronounced (Kruger et al. 2006) than those observed nowadays, when instead a general excess mortality of males is recorded. However, recent observations show that this gender gap is slightly decreasing (Mesle 2006).

In general, it has been observed that during the life course, the occurrence of certain diseases such as cardiovascular diseases, lung cancer and accidents, are more frequent among males (Guralnik et al. 2000) and this trend is confirmed when extending the comparison to the top 12 causes of deaths (Austad, 2006). Accordingly, disease-specific incidence rates may explain the observed sex differences in mortality patterns. The cumulative effect of these differences throughout the life course, results

⁴ GéDAP – Université Catholique de Louvain - Belgium

⁵ Department of Biomedical Sciences – Università degli Studi di Sassari - Italy

⁶ GéDAP – Université Catholique de Louvain – Belgium ; Università degli Studi di Cagliari - Italy

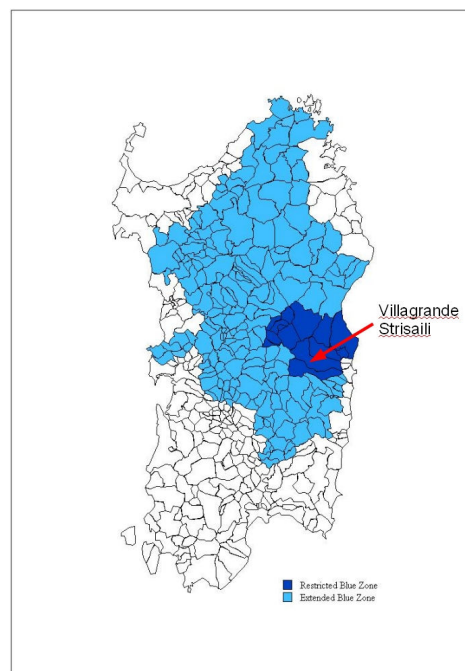
in a greater proportion of females surviving at more advanced ages. In populations experiencing a low mortality, the feminity ratio (F:M) among centenarians is usually above 4, i.e. there are more than 4 female centenarians for one male (*Human Mortality Database*). However, recent research have shown that in certain populations the sex ratio among the oldest olds may be remarkably lower, and when this occurs it is often the result of a reduced excess of male mortality rather than a higher mortality of women (Franceschi et al. 2000, Passarino et al. 2002, Robine et al. 2006). Among these populations, the one living in the Mediterranean island of Sardinia certainly represents an interesting case study. This region has been suspected to be characterized by a particularly high male longevity (Deiana et al. 1999). Data were carefully validated by using all available data sources in order to avoid cases of under-registration and age exaggeration (Poulain et al. 2006). Based on these validation results, an area with significantly higher levels of longevity and lower sex ratios among centenarians has been identified in the central-eastern part of the island, and was called the *Blue Zone* (Poulain et al. 2004).

This higher longevity *Blue Zone* encompasses nearly half of the 377 Sardinian municipalities, most of which are located in the mountainous region and includes one fourth of the whole Sardinian population (shown as extended *Blue Zone* on Figure 1). Based on the value of the 'Extreme Longevity Index' (ELI⁷), longevity of men born between 1881 and 1900 in the *Blue Zone* proved to be about twice higher than in the other part of Sardinia. In addition, a restricted group of 14 villages located in Ogliastra and Barbagia was identified according to their relative higher longevity level (shown as restricted *Blue Zone* on Figure 1). This more restricted zone, with about 42,000 inhabitants, shows an average ELI for males five times higher compared to the non *Blue zone* area. Moreover the feminity ratio among centenarians in this population is close to one, as 47 male centenarians were found and only 44 female

⁵ ELI (extreme longevity index) is the probability for a newborn in a given place to reach 100 wherever he or she lives at that time.

centenarians⁸ which means that the gender gap in mortality is inexistent in this population.

Figure 1 - Map of the extended and restricted Sardinian Blue Zone and localisation of Villagrande Strisaili.



From these findings, the following question arises: *How is it possible that in this area men live as long as women?* In order to investigate this question, an in-depth analysis of individual demographic data may be helpful. With this purpose, the population of the municipality of Villagrande Strisaili was selected, as it is the municipality with the highest ELI value in Sardinia (10.8 centenarians per 1,000 newborns) and the life trajectory of each individual born in the village during the period 1876–1915 was followed from birth to death.

⁸ In the extended *Blue Zone*, the same sex ratio was 1.35 compared to 2.43 in the rest of Sardinia.

The analysis of the collected data allows the comparison of the mortality trajectory for men and women. Moreover the life table estimates of Villagrande Strisaili have been compared with the corresponding ones for Italy and Sweden extracted from the *Human Mortality Database*⁹. The analysis of mortality trajectories also allows the identification in more detail of the critical age interval for determining population level differentials in longevity among the populations considered.

In the discussion, possible explanations for the lack of excess male mortality in the *Blue Zone* in Sardinia will be reviewed considering the existing literature.

Setting, data, and methods

The village of Villagrande Strisaili is located at 700 meters above sea level in the region of Ogliastra, but the altitude of its territory ranges from sea level to Punta La Marmora at 1,834 meters. 3,501 inhabitants were living in Villagrande on 1 January 2008 (ISTAT) and most of them are still involved in agropastoral activities and have maintained a lot of traditions in their daily life style.

The database developed for the present study includes all individuals born in Villagrande Strisaili from 1876 to 1915¹⁰. For each individual we traced the exact date at death or the confirmation of survival. The data was gathered from civil registers (which records all births, marriages and deaths), parish registers and population

⁹ Web-site: <http://www.mortality.org/>

¹⁰ Although the data are available since 1866, the year when the civil registers were first held in Sardinian municipalities, only the cohorts born after 1876 are considered, as the data related to the earlier birth cohorts are less complete. The last year of observation, 1915, corresponds to the availability of similar figures for Italy in the *Human Mortality Database*[®] that we used for comparative purposes. Accordingly, the cohorts of those who were at the time of investigation at least 95 years old are included.

registers (*anagrafe*). A total number of 2,142 newborns¹¹ have been considered in this study and these have been divided into four categories:

- newborns who died in Villagrande itself (1,753 newborns);
- newborns who died outside Villagrande (232 newborns);
- newborns who are still alive at the date of investigation (44 newborns)¹²;
- newborns for whom the date of death or the survival is unknown (113 newborns).

All information has been collected in the municipality population office itself and was cross-checked and integrated with data from parish registers. Furthermore, two additional data sources were also used: information reported in the military register and orally reported information from some still living relatives of these newborns.

With regard to newborns that died outside the village, the information was recovered by using the following different sources:

- annotations on date and place of death reported in the margin of the birth certificate;
- population registers called *anagrafe* (by family or individual) available since 1930;
- online electronic *anagrafe* available since 1980s.

In this way, we were able to reduce significantly the number of missing dates of death due to lack of information (113 newborns). For these newborns with missing date of death, we made the distinction between out-migrants and those where no emigration information was available. For the former group, the exact date of their

¹¹ From the totality of newborns in Villagrande were excluded only those babies (136 out of 2278) that clearly were accidentally born in the village, where their parents were living at that time for working reasons. To identify these newborns we used the origins of both parents and their occupation (miners, workers involved in the construction of roads and train networks, police officers) as usually reported in the birth certificate.

¹² As of 5 September 2006

departure from Villagrande was used as censoring date. For those without information on emigration, we used their marriage date or the latest appearance of their name in administrative documents as censoring date. Doing so, only for 27 newborns information was lacking on their survival, whereas for 86 others we found at least information attesting their survival until the censoring date.

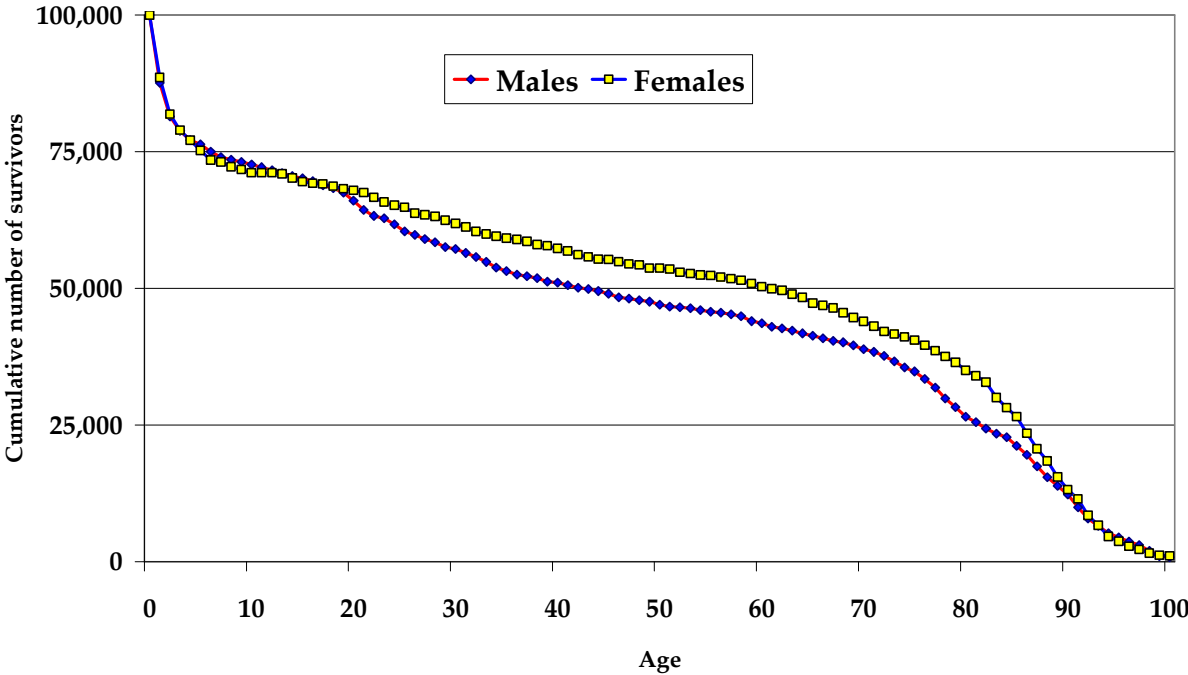
A final coverage of above 98% was reached – which must be considered exceptional in the context of historical demography and family reconstruction. In such a favourable situation, the exclusion of the 27 newborns without information and the censoring of the 86 other newborns cannot affect the validity of our findings.

For comparative purposes, data for Italy and Sweden have been collected from birth cohort life tables available in the *Human Mortality Database* for the same birth cohorts 1876-1915 considered for Villagrande Strisaili and involving separately males and females by single year of birth and age.

Results

The survival curves from birth till age 100 shown in figure 2 for all birth cohorts 1876-1915 were estimated by using the survival method separately for men and women and considering all birth cohorts together.

Figure 2. Comparing survival curves for males and females in Villagrande for a hypothetical cohort of 100,000 newborns of the years 1876-1915.



At age 5, one newborn out of five has already died and an additional tenth did not reach the age of 18. At 50 years old, 47% of males and 54% of females are surviving. Starting from that age – from 50 to 75 years old – males and females seem to be exposed to the same mortality risk. After 75 years, men record higher risks of dying compared to females but in the 80s the difference between males and females is inversed so that finally at age 90 about 13% of the newborns – both for males and

females - are surviving. That means that 1 newborn out of 8 reached 90 years old and that men live as long as women in Villagrande. The comparison with strictly comparable data for Italy and Sweden extracted from the *Human Mortality Database* is presented in figures 3a and 3b separately for men and women. The situation is really exceptional for Villagrande compared to these two populations and indicate that the mortality trajectories for men and women should be analysed more into detail. First, the measures of infant and child mortality may be compared with the ones recorded in Sardinia and in the whole of Italy for the same period. For Sardinia as a whole, two major demographic features emerge, underlying the peculiarity of this region with regard to the mortality levels at young ages: a lower infant mortality (0 to 2 years) and higher child mortality (2 to 5 years). During the first year of life the situation is even better in Villagrande, where infant mortality rates are 9.4% for girls and 10.8% for boys, while in Sardinia an average rate of 15.7% is recorded for the same period and 18.7% in Italy as a whole (Gatti 2002). Comparing rates for Villagrande newborns in all groups of ages up to 5 years with Sardinian figures (Coletti 1908), it is clear that the rates for Villagrande are consistently lower than in the whole of Sardinia, even if the rates show an increase after the age of 2 for Villagrande. Considering the mortality rates from age 5 until age 18, we observe that in Villagrande seven out of ten newborns, both for boys and girls, are surviving at age 18.

The analysis of adult mortality appears to be more complex since it is mainly due to external causes such as losses in WWI for men and maternal mortality for women. We estimated that a total of 40 males born in Villagrande between 1876 and 1915 died during WWI and that half of them were born in the period 1891-1895. For women of the same age group, maternal mortality was also high during the reproduction period. Orrù and Putzolu (1994), in their study on the diffusion of professional assistants for delivery, pointed out that in 1899 there was no professional assistant in Villagrande Strisaili and in the surrounding area. However,

in the village, a maternal mortality rate varying between 10,6 and 11,1 per 1,000 births was estimated (Salaris 2009), which is in line with the levels recorded in other Sardinian villages and in other European countries for the same period (Gatti 1999). Due to the higher mortality risk for both sexes between 18 and 50, only 50% of men and 55% of women are surviving at age 50 and these proportions are similar to the ones observed for the whole of Italy, while the Swedish ones are largely lower (Figures 3a and 3b).

About three out of four males and females surviving at age 50 finally reached age 75. When comparing these figures with the corresponding Italian and Swedish ones (Figure 4a), a lower mortality risk is evident mostly for men, even compared with Sweden, a population considered as one of the world leaders in longevity (Wilmoth et al. 1996). Accordingly, a crossover occurs for men aged 75 between the survival curves for Villagrande and Sweden, while for women the two curves join after 80 years old without crossover. Finally, it worth noting that the number of nonagenarians is almost evenly balanced between men and women, and this result must be considered exceptional compared with the situation in Italy and Sweden (Figure 4b).

Figure 3a. Comparison of survival curves for males in Villagrande with the corresponding data extracted from the Human Mortality database for the whole of Italy and Sweden (birth cohorts from 1876 till 1915 included).

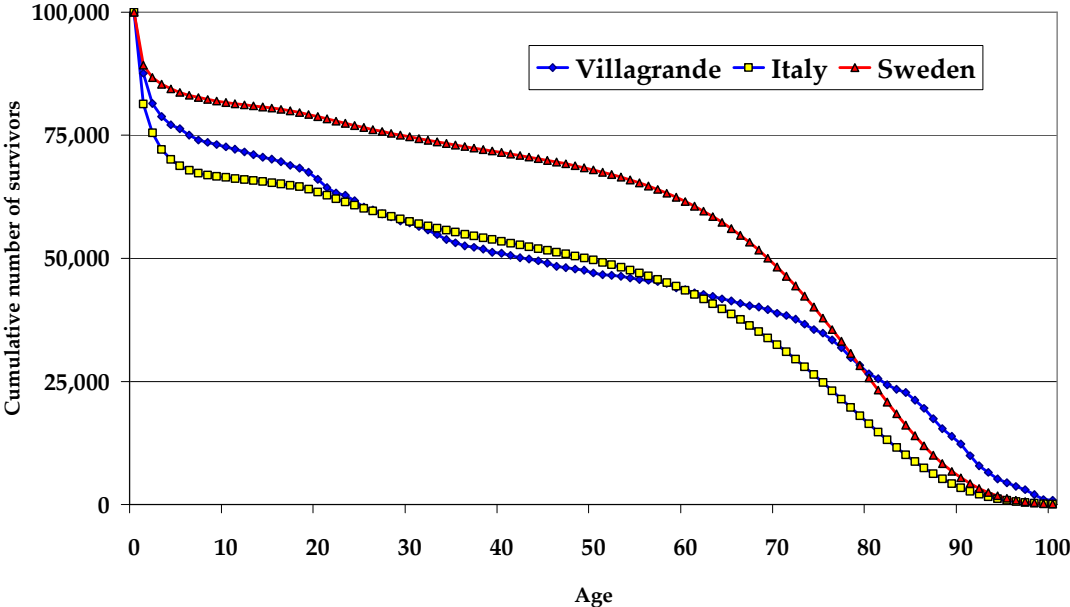


Figure 3b. Comparing survival for females in Villagrande with strictly comparable data extracted from the Human Mortality database for the whole of Italy and Sweden (birth cohorts from 1876 till 1915 included).

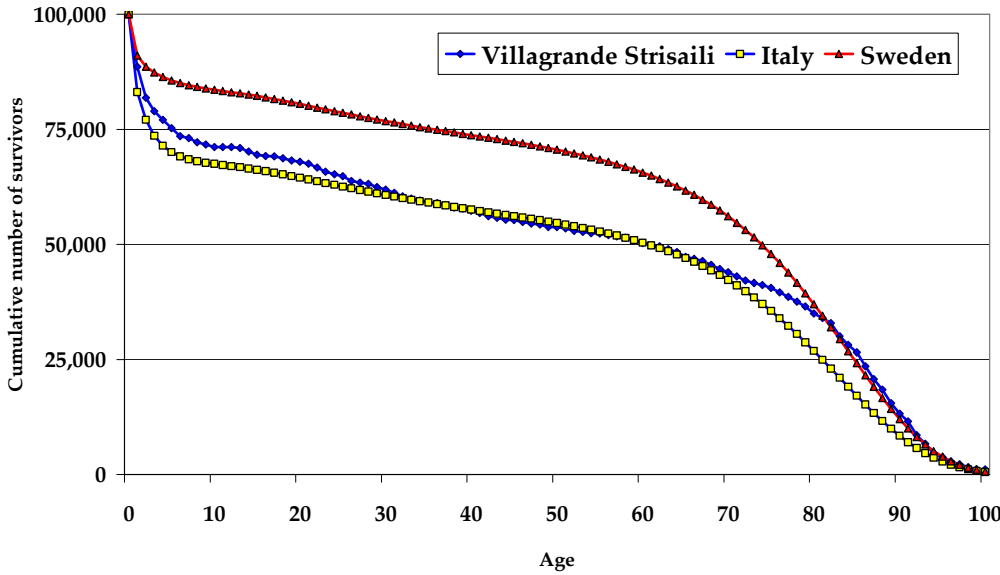


Figure 4a. Probability of surviving from 50 to 75 for Villagrande, Italy and Sweden (birth cohorts from 1876 till 1915 included).

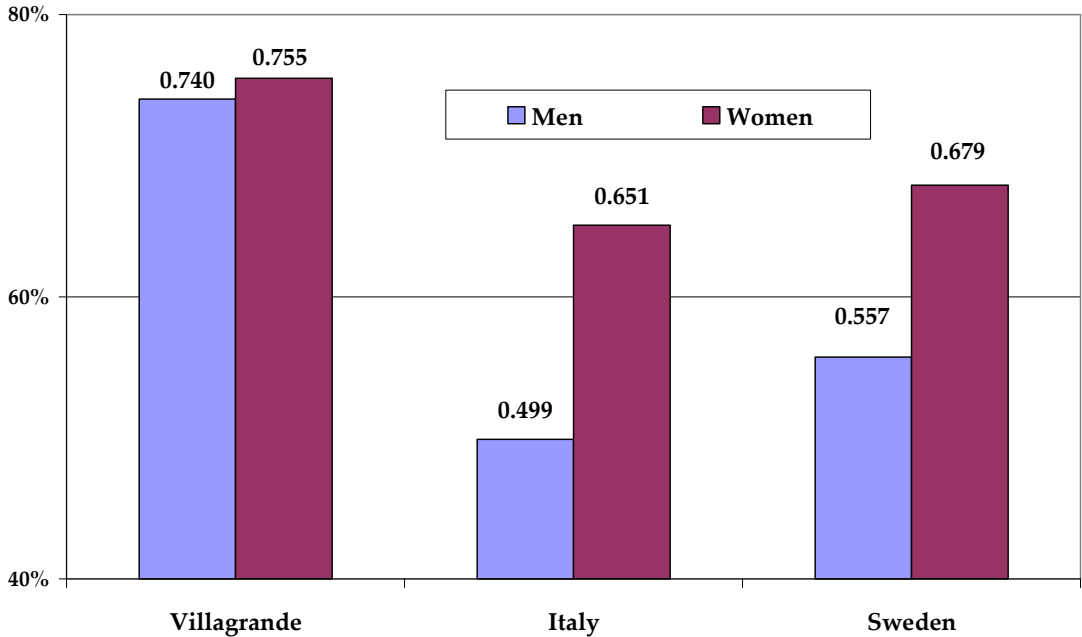
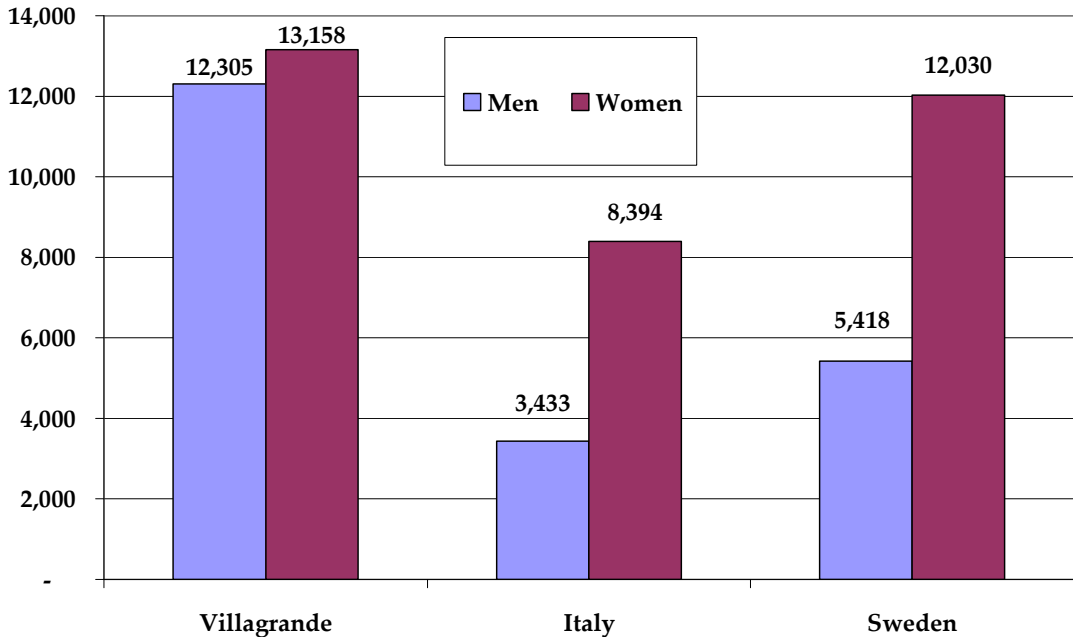


Figure 4b. Probability of reaching the age 90 in Villagrande, compared to Italy and Sweden (for 100,000 newborns in the life table).



Discussion

Why do men usually live less than women? This question has been addressed in several studies where potential explanatory factors are examined (Wingard 1984, Waldron 1985, Verbrugge et al. 1987, Antonini 1991, Crose 1997, Kalben 2000, Case et al. 2005, Austad 2006). Why men live as long as women in Villagrande and, more generally, in the Sardinian longevity *Blue Zone* is a related question emerging from the results presented here. To discuss this question more in detail, we will review some of the factors proposed in the literature to explain female longevity advantage, and try to understand why they seem not to work in the population under concern.

The first group of factors invoked to explain the female advantage in longevity relates to biology. Among these, the role of gender-specific genes has been investigated. Women have two X chromosomes, one from their father and the other from their mother, while males have only one X chromosome and one Y chromosome. Because of this, women have two cell lines, i.e. they have two different sources for some key genes. In females, if any allele detrimental for survival¹³ is inherited, the allele on the second X chromosome could compensate the expression of the first one, a protective strategy that is lacking in males. It is possible that in genetically homogenous populations such as Sardinians, the probability that for females both X chromosomes carry the same allele (homozygosity) is so high as to reduce the protective effect of having two X chromosomes. A different explanation would be that some recessive genes may be identified on the X chromosome that would be favourable for longevity and are expressed only in males hemizygotes. This may be the possible effect of G6PD deficiency, a X-linked disorder quite common in the island (Sanna et al. 1990), that was hypothesized to explain Sardinian longevity for males (Schwartz et al. 2004). Besides, G6PD deficiency was proven to be a protective factor against cardiovascular diseases (Meloni et al. 2008).

¹³ One of slightly more than 1,000 genes located on the X chromosome

Other genetic traits have been claimed to favour male longevity, such as the β -thalassemia trait that confers some “protection” against the development of premature cardiovascular diseases. A longitudinal study carried out in Northern Italy reported a significantly lower prevalence of β -thalassemia trait among myocardial infarction survivors (4.43% vs. 11.3%) (Gallerani et al. 1991). In addition both G6PD deficiency and β -thalassemia trait show a high prevalence in Sardinia, probably linked to the longstanding impact of endemic malaria up to the '40s (Luzzatto et al. 1995). Therefore it would be worthwhile to compare their different spatial patterns to that of longevity. Unfortunately, at the moment, the few available data do not support the existence of a higher prevalence of G6PD deficiency, β -thalassemia and past malaria in the Blue Zone what would contribute to explain a higher longevity for males in that region (Sanna et al. 1997).

The role of mitochondrial DNA on gender differential longevity has also been investigated. In particular, the frequency of a polymorphic variant called J haplogroup was reported to be increased in male centenarians from continental Italy (Debenedictis et al. 1999). In the Ogliastra population, the frequency of such haplogroup is more than twice that of Sardinia as a whole (18.3% vs. 6.7%) (Fraumene et al. 2003) making this polymorphism a valuable candidate to explain the absence of excess male mortality.

Other potential explanatory factors in this first group include anthropometric differences often observed between males and females. The difference in average height between male and female may be less important in the BZ population, while the average body mass index of females may be relatively higher compared to males. If confirmed, these two assumptions would lead to a longevity advantage for males as there is an inverse relationship between both height and body mass index and longevity (Samaras et al. 2003, Salaris et al. 2006). Evidence derived from military conscripts shows a shorter height on average for men in the Blue zone compared to

those in the whole of Sardinia (Salaris et al. 2006), whereas no such data are available for women, thus precluding the possibility to test this hypothesis.

Lastly, hormonal differences have also been associated with the gender gap in longevity mainly through their effects on cholesterol metabolism and endothelial function. For instance, low serum testosterone levels have been proved to be associated with increased risk of mortality in elderly men independently from the overall health status (Laughlin et al. 2008). The study of the endocrine profile of Sardinian oldest olds showed markedly higher testosterone serum levels and lower estradiol than in younger controls, although the real significance of these results is still to be clarified (Delitala et al. 2006).

The second group of factors is related to individual behaviour and socio-cultural traits of the population. They include possible differences in nutrition between males and females, but also in physical activity and energy expenditure. Such differences in behaviour are mostly linked to the different occupations of males and females as well as other aspects of their life style. Although no specific study up to now ascertained any gender-related differences in Sardinian nutrition, it may be hypothesized that the distinct role of women within the families and the prevalent occupation of men may have induced appreciable differences in dietary patterns of both genders. Higher wine consumption by men was also claimed to have a positive effect on longevity (Corder et al. 2006). Moreover, as in the longevity *Blue Zone* men were mostly shepherds or farmers, they likely did more physical activity than women did during their active life. The additional circumstance of living in a mountainous area increases the daily energy expenditure of most adult men compared to adult women, also favouring longevity (Warburton et al. 2006). Some specific character traits of males compared to females like less stress, more optimism, and a better ability to take more quickly advantage of the positive aspects of the health transition, may also play a positive role for men. The absence of a gender gap

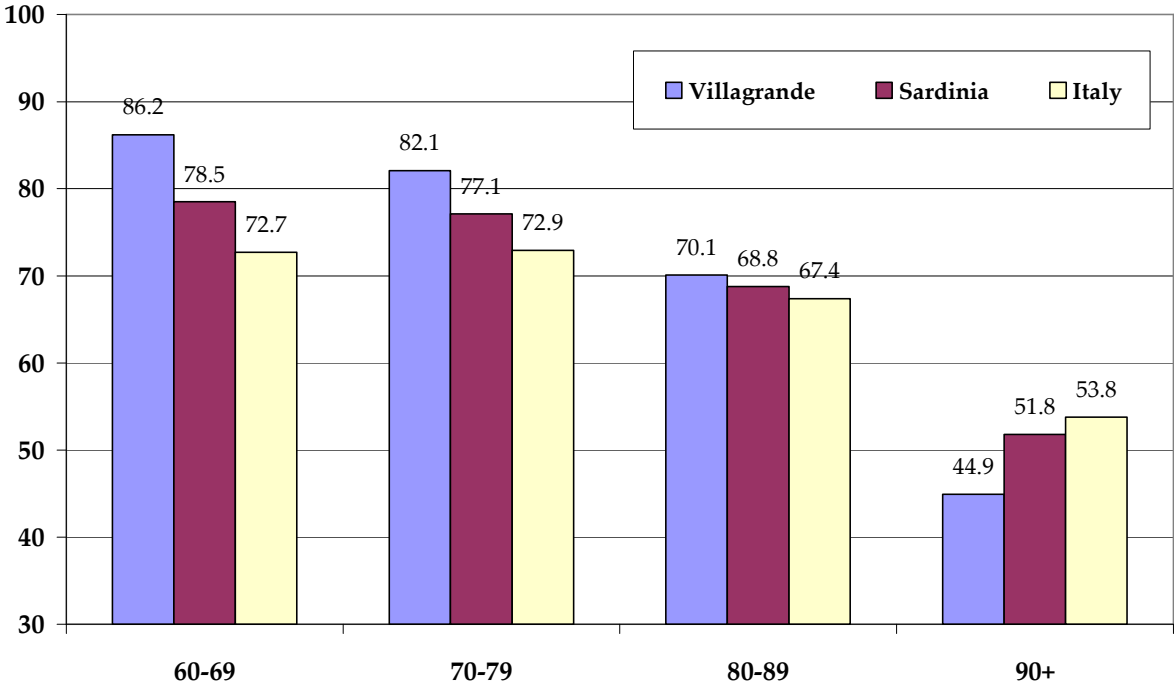
in mortality risks might also be attributed to differences in men and women social behaviour within a rather archaic society that acknowledges distinct roles to males and females. In particular, the population of central Sardinia during the centuries experienced a kind of matriarchal organisation (Pitzalis-Acciaro 1978) that lasted till the onset of industrialisation (late 1950s). Males, farmers and shepherds, were the workforce of the community and spent a great part of their active life outside, while females were the trustees of transmission of the ethical values, including self-defence, implying a certain degree of aggressivity, a strong sense of honour and the care of goods and property. This specific role of women within the family, and hence within the community, probably implied an increased 'chronic stress burden' compared to men, which ultimately may have contributed to increase female mortality (Oppo 1990).

Some specific socio-cultural traits of the population living in the *Blue Zone* may also favour male longevity. It is well established that a larger family support and some living arrangements like living in a married couple, may help to live longer (Gaymu et al. 2008). So, if men married younger women, they will largely escape widowhood that is considered as negative for living longer, at least for men (Foster et al. 1984). Figure 5 shows that the proportion of married males among oldest olds is larger in Villagrande compared to the situation in the whole of Sardinia and in Italy. Moreover, a high level of remarriage for men increases the age difference with their last spouse. Among the 324 married men found in our database, who died above 80, 41 (13%) were married more than once and the age difference with their (last) spouse was for 136 of them (39%) larger than 10 years (the median is 9.1) and only 25 (7%) had a spouse who was older. Such larger age differences between spouses is favourable for male longevity. In addition, in presence of a more favourable family environment, people may live longer. In Sardinia, older women tend to live more often alone after widowhood while older men will live preferably with an unmarried child. This situation also favours male longevity.

Also the possible role of fertility, including the timing of the reproductive period, on women’s survival has to be considered. Historically, Sardinian women were characterized by high fertility and a high age at birth of their children (Bernardi et al. 2007). This, on the one hand, may have increased the susceptibility to diseases of the mothers associated with pregnancy, and on the other hand may have reduced the effective exposure to levels of estrogen which can protect against cardiovascular diseases. Moreover, a reduced duration of the fertile period could have diminished the protective effect of continued loss of iron due to the menstrual cycle.

Finally, some gender stereotypes and related social norms may favour men compared to women and this seems to be the case in the longevity *Blue Zone*, where oldest men are the target of intense attention within the family but also in the local community; this may bring them support for living longer.

Figure 5. Comparison of the relative proportion of married men for oldest olds in the male population of Villagrande, Sardinia and Italy in 2008 (Source: ISTAT).



CONCLUSION

An intensive data collection and complete family reconstruction allow finding the date of death or confirming the survival at the date of survey of almost all 2142 children born in Villagrande during the years 1876-1915. The cohort life table has been estimated and the mortality trajectory identified separately for men and women. The survival curves have been compared with similar ones for Italy and Sweden. The exceptional situation of men in Villagrande emerges from that comparison, as men are living as long as women do, which is usual. Three conclusions emerge from our investigations. First, among the possible reasons explaining why men apparently live as long as women in Villagrande and in the Sardinian *Blue Zone*, age exaggeration for men has to be clearly excluded as a result of the age validation exercise. Secondly, it is highly probable that no single explanation can be found for justifying such an exceptional situation. Thirdly, we consider that all the above mentioned factors may work jointly, so that apparently the usually found paradox of lower morbidity and higher mortality for men is not valid for the population of the longevity *Blue Zone*. In conclusion, the research should be continued keeping in mind that only a multidisciplinary approach will help finding the underlying set of explanatory factors.

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Annex: Comparable survival figures for Villagrande, Italy and Sweden
Comparison of survival curves for males in Villagrande with the corresponding data extracted from the Human Mortality database for the whole Italy and Sweden (birth cohorts from 1876 till 1915 included).

	M A L E S			F E M A L E S		
	Villagrande	Italy	Sweden	Villagrande	Italy	Sweden
0	100000	100000	100000	100000	100000	100000
5	76309	68773	83680	75244	70054	85629
10	72635	66432	81661	71150	67492	83574
15	70156	65357	80510	69493	66206	82264
20	66024	63476	78800	67934	64487	80509
25	60422	60164	76563	64815	62552	78589
30	57208	57513	74664	61891	60736	76793
35	53168	55298	73018	59162	59064	75202
40	51056	53435	71489	57310	57555	73740
45	49036	51640	69909	55263	56132	72277
50	47016	49639	67969	53704	54646	70579
55	45730	47042	65329	52339	52798	68425
60	43618	43515	61556	50292	50378	65624
65	41322	38707	56048	47271	47008	61719
70	38843	32455	48233	43957	42252	56080
75	34803	24775	37885	40546	35561	47924
80	26538	16374	25704	34990	26824	36970
85	21212	8729	13960	26511	17088	24166
90	12305	3433	5418	13158	8394	12030
95	4408	870	1266	3704	2785	3853
100	826	120	145	1072	520	636