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Family clustering in Sardinian longevity: A genealogical approach

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Abstract

This paper aims to discuss the validation and family determinants affecting the longevity of Sardinian centenarians, using a genealogical approach. This preliminary study presents the first results of a genealogical tree reconstruction of selected centenarians aged 105 and over, from certain areas. These are mostly situated in the province of Nuoro, an area with the highest rate of centenarians and where the female-to-male sex ratio tends to be male-biased. An accurate centenarian age validation was performed that required a meticulous examination of numerous civil status records and parish registers. An important finding was that longevity occurs among the ascendants of a particular branch of the family. The data used are still provisional but, should it apply to other validated cases, it would provide empirical evidence of a genetic component in longevity. A more thorough examination of the data available may yield deeper insights into the role played by endogamy and consanguinity.

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1. Introduction

The oldest old have recently enjoyed steady growth in life expectancy. The number of centenarians has approximately been doubling every 10 years since 1960 as a result of the growing numbers of over 80-year-olds in the population (Vaupel and Jeune, 1995; Wilmoth et al., 2000; Robine and Caselli, 2005). Centenarians appear to be a unique model to investigate the biological and non-biological determinants of aging and longevity as well as how these intertwine (Baggio et al., 1998).

This paper is part of the multidisciplinary and multipurpose research project, "A demographic analysis

* Corresponding author. *E-mail address:* graziella.caselli@uniroma1.it (G. Caselli). of Sardinian Longevity-AKEa2".¹ It aims to document and verify the remarkable pattern of low mortality among 80 and over year-old Sardinian males and to explore the underlying reasons (Caselli and Lipsi, 2005). A large number of exceptionally old men (aged 100 and over or 105 and over) live in Sardinia and the sex ratio among centenarians is unexpectedly low, particularly in the inland municipalities of Nuoro province (Poulain et al., 2004).

The Sardinian population of the 2001 census (October 21) was roughly 1,631,880, including 187 centenarians (53

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¹ AKeA 2 is the acronym of the survey on Sardinian Centenarians, directed by James Vaupel, Max Planck Institute for Demographic Research, Rostock, Germany ("Demographic Analysis of Longevity. A Survey on Sardinian Centenarians", financed by NIA, US, subcontract with Duke University n.03-SC-NIH-1027). The survey follows a previous research project on Sardinian centenarians, named AKeA1.

men and 134 women), with a ratio of 1.2 centenarians per 10,000 inhabitants (compared to 1.0 for Italy) and a centenarian female-to-male sex ratio of 2.5:1. Elsewhere in Italy, the centenarian sex ratio is slightly over 5. In other low mortality countries, it ranges from 5 to 7 (Robine and Caselli, 2005). In certain pockets of Sardinia, the female sex ratio is even lower, in some areas it hovers only slightly above 1 (Poulain et al., 2004).

The demographic history of Sardinia is striking. The cohorts born at the turn of the 20th century, i.e., the cohorts subject to this study, experienced a particularly interesting life-course.

They saw the lowest infant (and especially neonatal) mortality rates recorded in Italy at that time and stillbirth rates lower than the national average (Pozzi, 2000) even though the maternal mean age at birth was particularly high. However, after age 2, the risk of dying during childhood and youth was substantially higher for Sardinian cohorts compared to the corresponding cohorts from other Italian regions. Exposure to malaria, which was still endemic on the island at that time, cannot be ignored and deserves further attention.

Genetic selection has occurred over the centuries owing to malaria. This selection can provoke glucose-6-phosphate deidrogenase (G6PD) deficiency, which is double as high for Sardinian centenarians as it is for the rest of the population. Recent research claims that the lack of this enzyme contributes to longevity in Sardinia and to the exceptionally high number of male centenarians (Deiana et al., in press). Preliminary results seem to indicate that this mutation is present in 11.2% female and 25% male centenarians.²

Hence, generations of centenarians have been able to tap into a great vital potential. Struck down by malaria at young ages, the usual selection process was triggered with the elimination of the relatively fragile and the emergence of individuals more robust due to these genetic mutations.

Sardinia has often been the focus of genetic studies as the population exhibits a number of unique features due to their isolation over the centuries and because they stem from a single familial stock (Cavalli Sforza et al., 2004).

It is also interesting that the men from these cohorts were engaged in the First World War, thus experiencing higher probabilities of death (compared to neighboring cohorts not directly involved in the war). Higher male mortality persisted for many years after the conflict ended, as recent research has pointed out. Having undergone further selection due to high mortality at young-adult ages, these cohorts may contain a large number of individuals at the threshold of old age who are particularly robust (Barbi and Caselli, 2003). Sardinian centenarians are an exceptionally homogeneous group in terms of socio-occupational status, with the men usually having been either peasants or shepherds, and the women housewives, following an age-old tradition handed down from parent to child. In view of the role the socio-occupational variable plays in mortality studies, this is not a discriminatory factor in the study of Sardinian centenarians.

The centenarians exhibited low mobility between their place of birth and current residence. Besides, a relatively high number of centenarians, particularly those aged 105 and over, lived in certain inland areas where environmental risk factors are lower than along the coast. The Italian Ministry of the Environment has listed 691 polluted sites in Sardinia located around the Gulfs of Cagliari and Oristano, in the extreme north-west of Sassari, in the area known as Porto Torres and along the north-east coast of Nuoro.

On Sardinia and particularly inland there is a considerable variation in the frequency of given surnames in different areas of the region (Zei et al., 1986) and in the prevalence of specific territorial surnames, assuming the practice of endogamy in the past. The literature has reported higher infant and child mortality among births from consanguineous marriage (Cavalli Sforza et al., 2004). Thus, we should be surprised to find so many Sardinian centenarians in highly endogamic communities? Does consanguinity generate a selection effect? Do individuals born to an endogamically selected couple have a longer life potential?

These and other considerations led us to believe that Sardinian centenarians are an ideal population group to test the impact of different determinants on survivorship to exceptionally old ages.

Longevity is, broadly speaking, the outcome of various factors of a genetic or environmental nature (early life factors, life style, education, occupation, place of residence, dietary habits, environment, etc.).³ The number of centenarians in developed societies is a fast growing segment of the population and has inspired research into the role played by genetics in longevity. The wide debate on many of the most relevant issues regarding familial longevity continues to date (Bouquet-Appel and Jakobi, 1991). Some authors are playing down the role of genetics in familial longevity and find that familial resemblance plays a minor role to the benefit of social explanations. An impressive and coherent series of epidemiological data in different populations (New England Americans, Mormons, Askenazi Jewsh, Islandic, Okinawan Japanese) indicate the presence of a strong familial and genetic component of longevity (Gudmundsson et al., 2000; Kerber et al., 2001; Perls et al., 2002; Terry et al., 2003; Terry et al., 2004; Atzmon et al., 2006; Schoenmaker et al., 2006; Willcox et al., 2006). These

² Preliminary results included in the AKeA2 Project on Demographic Analysis of Sardinian Longevity.

³ Hereafter, socio-economic, environmental, behavioral, and cultural factors will be referred to as the "environmental impact".

studies demonstrate that parents, siblings and the offspring of long-lived subjects, but not the spouses of the long-lived subjects who shared with them most part of their adult life, have a significant survival advantage, a higher probability to have been or to become long-living and a lower risk regarding the most important age-related diseases, such as cardio- and cerebro-vascular diseases, diabetes and cancer, when compared to the appropriate controls. Thus, longevity is present in many generations of the same families in spite of the great variations in life style and life expectancy of the last century. Moreover, the most recent studies on twins strongly support that genetic effects are important for survival at older ages, and that genetic influences on lifespan are minimal prior to age 60 (Hjelmborg et al., 2006).

Extreme longevity in a centenarian's family history supports the inheritability of oldest old longevity (Perls et al., 2000). This would imply increased homogeneity among the oldest old and could depend on shared genetic factors. Indeed, recent findings indicate that increased homozygosity at loci involved in human longevity occurs, suggesting two copies of the same allele may favor survival at later ages (Bonafe et al., 2001; Cardelli et al., 2006), this phenomenon likely being more frequent in consanguineous subjects, assuming that such alleles are recessive.

Demography could help to provide deeper insights into the role inheritability plays in longevity, even in the absence of genetic variables. To this end, data are needed to reconstruct the family histories of the oldest old and, in this instance, of centenarians from a distinct geographical area. Next, an analysis of genealogies and family life histories can be performed, using features from individual trajectories to reconstruct the life history of each sibling.

The paper first aims to illustrate the various steps taken to reconstruct the genealogies of centenarians aged 105 and over, using a thorough validation procedure, combining civil and parish data. Individuals aged 105–109 and 110 and over are named "semi-supercentenarians" and "supercentenarians", respectively. The second aim is to look into the family relationships and the family clustering of Sardinian longevity, using the genealogical trees of the study subjects. To date, 11 genealogical trees have been built. This paper presents the initial results of an ongoing research that should be extended in the future.

2. Materials

Our sample consists of 207 centenarians (100 men and 107 women) and certainly is representative of a population that only some years previously numbered 310 centenarians (January 1, 1997 to September 15, 2003). The centenarians participating in the AKeA2 project were selected on the basis of their place of birth. Only a small proportion of the centenarian data collected will be used in our study, however. Selected genealogies are examined that typify certain traits of Sardinian centenarians and that highlight the wealth of the material available.

A total of 53 persons aged 105 and over (17% of the total number of centenarians) from 44 municipalities on

Table 1

| Centenarians (aged 105 and over) by sex and age, 1997–200 | Centenarians | (aged 10 | 5 and over |) by sex and a | ge, 1997–2003 |
|---|--------------|----------|------------|----------------|---------------|
|---|--------------|----------|------------|----------------|---------------|

| Provinces, region | 105–107 | 108–109 | 110 and over | Total 105 and over | Prov./reg per 100 | Total population, 2001 census | 105 and over population per 10,000 |
|-------------------|---------|---------|--------------------|--------------------|-------------------|-------------------------------|------------------------------------|
| Men | | | | | | | |
| Cagliari | 3 | 2 | _ | 5 | 25.0 | 371,528 | 0.13 |
| Nuoro | 7 | 1 | 2 | 10 | 50.0 | 130,223 | 0.77 |
| Oristano | 2 | _ | | 2 | 10.0 | 75,296 | 0.27 |
| Sassari | 2 | 1 | _ | 3 | 15.0 | 222,191 | 0.14 |
| Sardinia | 14 | 4 | 2 | 20 | 100.0 | 799,238 | 0.25 |
| Women | | | | | | | |
| Cagliari | 11 | 2 | _ | 13 | 39.4 | 388,783 | 0.33 |
| Nuoro | 6 | 3 | _ | 9 | 27.3 | 134,636 | 0.67 |
| Oristano | 2 | 1 | _ | 3 | 9.1 | 77,786 | 0.39 |
| Sassari | 8 | _ | _ | 8 | 24.1 | 231,437 | 0.35 |
| Sardinia | 27 | 6 | _ | 33 | 100.0 | 832,642 | 0.41 |
| Men and women | | | | | | | |
| Cagliari | 14 | 4 | _ | 18 (15) | 34.0 | 760,311 | 0.24 |
| Nuoro | 13 | 4 | 2 | 19 (16) | 35.8 | 264,859 | 0.72 |
| Oristano | 4 | 1 | _ | 5 (5) | 9.4 | 153,082 | 0.33 |
| Sassari | 10 | 1 | _ | 11 (8) | 20.8 | 453,628 | 0.24 |
| Sardinia | 41 | 10 | 2 | 53 (44) | 100.0 | 1,631,880 | 0.32 |
| Female/male ratio | 5 | | | | | | |
| Cagliari | 3.7 | _ | 1.0 ^a | 2.6 | _ | _ | _ |
| Nuoro | 0.9 | _ | 1.0^{a} | 0.9 | _ | _ | _ |
| Oristano | 1.0 | - | _ | 1.5 | _ | _ | _ |
| Sassari | 4.0 | - | _ | 2.7 | _ | _ | _ |
| Sardinia | 1.9 | _ | 1.0 ^a | 1.7 | _ | _ | _ |

Total population by sex at 2001 census, proportion of centenarians, and F/M ratios. Provinces and Sardinia.

At age 108 and over.N.B. The number of municipalities is given in brackets. Source:www.istat..it and AKeA2 project.

the island were identified from the 1997–2003 centenarian survey. Of these, 44 have been validated, with no age errors occurring and no discrepancy between the corresponding vital records and parish registers for the 11 genealogical trees reconstructed.

Of the 53 centenarians, 33 are women and 20 are men. In the province of Cagliari, centenarian women number 39.4% of the total centenarian population, whereas only 9.1% in the province of Oristano (Table 1). The distribution of centenarian men differs from province to province: 50% live in Nuoro, 25% in Cagliari, and the rest in Sassari and Oristano. The proportion of those aged 105 and over in Nuoro on the total population is three times that of the whole region. For women, it is less than double. Since the distribution of men and women is different, the sex ratio varies between the four provinces, too. (see Table 1).

Of particular note are two men aged 110 and over who live in the province of Nuoro.

2.1. Validation process

The validation process began some years ago as part of the preliminary AKeA1 study. A total of 141 centenarians were validated in the AKeA1 study, the number has risen to 207 centenarians in AKeA 2; here, only some of the centenarians validated in the first survey were included. For these, further details were collected from a larger series of documents generated by the fact that AKeA 2 addressed other aspects, such as endogamy, the fertility of centenarian women (age at first birth, birth intervals, the number of children, etc.), and the survivorship of the centenarians' descendants.

As mentioned above, *civil records and parish registers* were used for the validation process and to build genealogical trees. The *civil records* contain birth, death, and marriage registers held in each municipality and date from 1866. Local population registers kept on a continuous basis in each municipality, i.e., *Anagrafe*, were used, too. For complete age validation, the following documents were looked at: the birth certificate, marriage certificate(s), the *Anagrafe* sheet, the birth certificate of both parents as well as their marriage and death certificates, the birth certificates of all children of the centenarian in question and their *Anagrafe* sheets, and the birth certificates of the centenarian's siblings and their *Anagrafe* sheets. Complete validation is achieved only when the data are cross checked and proves to be consistent.

For the years before 1866, the following parish registers were used: "Libri dei battesimi" (baptismal registers), "Libri dei matrimony" (marriage registers), "Libri dei defunti" (burial registers), and "Status Animarum" (a sort of parish population censuses).

Besides providing additional elements to confirm age validation, these sources are useful to reconstruct the biographies of the centenarians' ancestors. Data are available on the past history of the family of origin, their parents, grandparents, great-great-grandparents, and siblings. Often, the data also regard the extended family, uncles, aunts, grand uncles, grand aunts, etc. Owing to the availability of data from fairly wide-ranging and reliable samples and sources, family genealogies are reconstructed that throw light on longevity and inheritability, on the impact of the male or female line as well as on the role of vital events during early infancy. For Sardinian centenarians, data can also be traced on the composition and size of the family in question, for example, the place of birth and residence, the birth order and birth intervals, the month of birth, and the age of the parents at birth. These aspects have been shown to be of crucial importance in longevity studies: there may be, for instance, a link not only between the mother's age, but also between that of the father at birth and the survival of the child.

3. Results

As mentioned above, 11 genealogies are complete from the end of the 18th century: six for men and five for women. Of the 11 centenarians, three were born in the province of Cagliari, six in the province of Nuoro, and two in the province of Sassari. The priority selection criteria was age. Naturally, we began with the two individuals aged 110 and over, both males and both born in the Nuoro Province: Antono Todde in Tiana and Giovanni Efisio Emanuele Frau in Orroli. We applied the same criteria to women; the oldest woman selected was aged 109 years and lived in Cagliari. Priority was accorded to those linked by kinship. Thus, we have two cousins from Padria (Sassari province), one of whom, Antonietta Sebastiana Serra, died aged 106. The other, Caterina Serra, died on March 4, 2005, aged 107.

We have selected three family genealogies. Before homing in on them, let us first present some results that corroborate and further enrich the wealth of data available for Sardinia on endogamy, exogamy, and consanguinity. The communities where the centenarians lived were very isolated from the surrounding world. This was a common feature in Sardinia until recently and subject to research by anthropologists, geneticists, and demographers (Cavalli Sforza et al., 2004; Moroni et al., 1972; Caselli and Lipsi, 2005).

In view of the possible impact isolation has on the genetic structure and demography of the two first municipalities (Padria in the province of Sassari, Orroli in the province of Nuoro), parish marriage records were examined to identify married names, the place of residence, and any clerical dispensations because of kindred, for two 25-year periods (1856–1880⁴ e 1881–1905), so as to cover more or less two generations.

Preliminary analyses of the *parish registers* show that endogamy and intermarriage are common to both

 $^{^4}$ We excluded the years 1866–72 for Padria as the records were suspended by the bishop.

municipalities and that both marriage partners are more often than not from the same municipality. "Non-local"⁵ partners are usually from neighboring parishes, merely a few kilometres away. Moroni et al. (1972) found that between roughly 1850 and 1918 there was an increase in consanguineous marriages and the marriage distance between the partners' residences narrowed.

Endogamic marriages were quite frequent in Orroli until recently (90% in the first period analyzed, and 87% in the second). The figures are lower for Padria, but still high at 82% between 1856 and 1880, and 87% between 1881 and 1905.

We can not exclude possible oversights on part of the parish priest when he recorded the place of origin of the "non-local" partner. This applies to Padria, where the greater variety of surnames among couples implies a greater number of partners from outside.⁶

There are four to five partners with the same surname in Padria, and an average of eight in Orroli. New surnames from "outside" the area are more frequent in Padria.

To measure precisely the extent of a municipality's isolation, consanguinity was analyzed using church marriage dispensations for close degree kindred. This was high for both municipalities but the time trends were different: in Orroli, 10.5% of all (338) marriages between 1856 and 1880 were consanguineous, rising to 16.3% (of 418 marriages) between 1881 and 1905. The figures for Padria are 16.7% (of 281 marriages) and 11.6% (of 327), respectively.

The initial results on consanguinity are contradictory. To clarify the situation, a longer time span is needed and the analysis needs to be extended to include other communities. Some indirect measure of consanguinity, such as marital isonomy (both random and not random), should also be calculated.

When the source is reliable, marriage dispensations allow a precise and accurate measure of consanguinity. A drawback, however, is that the analysis only concerns marriages with the degree of kinship known, ignoring cases of distant kin relations, the latter which should not be overlooked in view of the genetic impact.

3.1. Three genealogies under focus: Antonio Todde's genealogical tree

The law of small numbers often does not support the hypotheses made but points into another direction. For this reason, we have decided to present three familial genealogies that are representative of three different contexts. One key factor is common to all four centenarians: each of them were related to someone who had reached age 100, either a family member or kin. Below, we present the Todde and Frau genealogical trees in Figs. 1 and 2 (the family refused authorization to publish the Serra genealogical tree).

There are a number of reasons underlying our decision to present the family tree of Antonio Todde, born in Tiana village in 1889 (on January 22). He died on January 3, 2002 aged 112 years and 346 days. The AKeA1 study certified that he was the oldest man in the world and was accepted as such by the Guinness Book of World Records 2002. He was the subject of a previous study assessing various aspects of his biography, health, and genetics (Deiana et al., 2002). Our decision to chose him was further due to the fact that he belonged to a family from a geographical zone with the highest number of known centenarians (Deiana et al., 1999).

Retracing the Todde's family tree was aided by the availability of documentary sources free of significant time gaps.

Civil status registers for the municipality of Tiana are complete for three series (births, marriages, and deaths) from the year 1866 onwards. They are well preserved, accessible, written in Italian, and easy to read. For life events prior to 1866, we used original parish registers for Tiana, stored at the Historical Archive of the Diocese of Oristano, again easily accessible, written in Castilian until 1820, then in Latin or Italian, and in Italian only after 1840.

The fact that the family comes from a small community, historically isolated with respect to the surrounding community, facilitated drawing up the family tree. This enabled us to trace back five generations, covering a time span from the last 20 years of the 18th century to date. Antonio Todde's family tree is presented in Fig. 1 and shows the age at death of each of the "relatives" for whom these data are available. Note that we made a distinction between cases with data based on a linkage between birth and/or baptism certificates and death and/or burial certificates, and those where the age was taken from death or burial registers. With regards to the latter, some approximation may have occurred. Experience, however, has proven that the ages contained in the civil registers are usually fairly accurate, more so than are parish registers, particularly more ancient ones.

Information was obtained by putting together the data contained in the following records:

- The baptism certificate, dated January 23, 1889 (only the date of the baptism is shown). The certificate contains the names and surnames of his parents, Francesco Maria Todde and Francesca Angela Deiana.
- The birth certificate⁷ from the civil status register, dated January, 26 1889 and referring to January 22, 1889 as the date of birth. The certificate includes the name of the

⁵ Marriages were usually celebrated in the bride's parish confirmed by the fact that the "non-local" is usually the groom that somewhat detracts from the results obtained. For a better overview of marriage exchanges, neighboring parishes should be included in the analysis or we need a nominative source, such as a census or a "status animarum" Unfortunately, these sources are not available for our communities.

⁶ Nonetheless, parish priests were usually very careful in filling out the marriage certificate, clearly stating the groom's origins and the parish of birth and residence when these did not coincide.

⁷ Birth certificates include a column next to the main text. This contains observations and margin notes and is especially useful as it provides information on the individual referred to in the certificate. Mention is made to events following birth, namely marriage and death, with dates recorded as well as the number of the certificate and the series it belongs to, thus facilitating linking between the various documents.

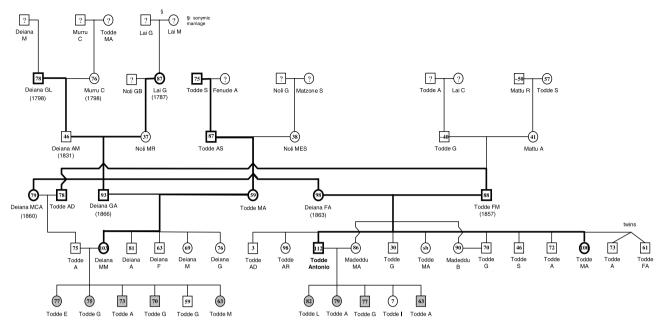


Fig. 1. Family tree of Antonio Todde (1889–2002). The number inside the symbols (circle for women and square for men) represents the age in years at death (symbols white) and the age reached (symbols grey).

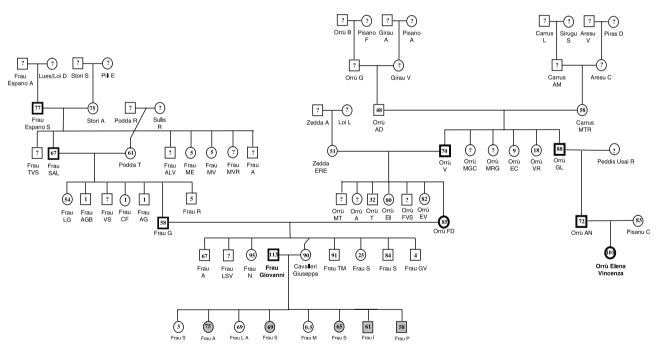


Fig. 2. Family tree of Giovanni Efisio Emanuele Frau (1890–2003). The number inside the symbols (circle for women and square for men) represents the age in years at death (symbols white) and the age reached (symbols grey).

father, whereas that of the mother is not listed. This is because both parents at that date had contracted a religious marriage only. Traditionally, the religious act of marriage was accorded higher priority, often resulting in a delay in the civil marriage ceremony, a phenomenon that has also been noted in other Italian regions of the time.

• The civil marriage certificate for Todde's parents. The marriage was celebrated on December 30, 1908, 25 years after the religious ceremony. The certificate contains the

legitimization of children born to them in marriage and still alive.

• The civil marriage certificate of Antonio Todde with Maria Antonia Madeddu, with the marriage dated September 15, 1920. The groom's age (31 years) is consistent with the date of birth, January 22, 1889. The paternity of the groom is correctly reported. Information regarding the marriage, with consistent dates, is provided in the margins of Antonio Todde's birth certificate.

- The birth certificates of five of the children born to the couple Todde Madeddu, spanning the years 1921–1933. In each of the certificates, the age of the father is consistent with that provided in the birth certificate.
- The death certificate of Antonio Todde, reporting his death on January 3, 2002. The information is consistent with the content provided in the margins of Antonio Todde's birth certificate.
- The same procedure was followed for the other members of the family, although in some cases (particularly for the older generations) fewer documents were found than for Antonio Todde.

An analysis of the family tree shows several cases of longevity among the first degree relatives of Antonio Todde. His father died at the age of 88 and his mother at the ripe old age of 98. One sister died at the age of 98 and another is still alive and aged 101 years (as of December 21, 2005).

Another sister died immediately after birth. A brother, the first-born, died before he reached age four. Considering that two other siblings died from violent causes at a youngadult age, overall the family would appear to enjoy unusual longevity.

In Sardinia and other populations, the names of family members are recorded in a family tree. According to custom, if a child died, then the name of the deceased child is given to another child born later. Hence, the family tree of Antonio Todde shows that the name of his fifth sister, Maria Agostina, who had died, was given to another sister born later and still alive, his ninth sister.

Of Antonio Todde's five children, four are still alive today, aged between 70 and 82. The only child who died during childhood (21 months old) was a girl. According to the family's recollection, her death was due to a form of anaemia, possibly talassemia, an illness that is frequently found in Sardinia. This assumption is plausible given that her father was a carrier of anaemia, as shown by molecular tests (Deiana et al., 2002).

The AKeA 1 study identified another centenarian who lived in Tiana and died there at the age of 103. She later turned out to be a cousin of Antonio Todde on his mother's side. This led to a further reconstruction of the maternal branch of Antonio Todde's family tree in that the cousin's ancestors were included.

The ages at death of Antonio Todde's grandparents and great-grandparents do not yield any exceptional cases of longevity among his paternal ancestors.

Among his mother's ancestors and collaterals, however, at least one instance of longevity is found, a brother, the father of the centenarian mentioned above. He reached 93 years of age. The maternal grandfather of his mother died in 1924, aged 87 years. Continuing with the maternal side, the great-grandfather, the great-grandmother, and the great-grandfather died aged 75, 87 (1874), and 75 (1873), respectively – all considered ripe ages at that time.

A close look at the family tree shows that endogamic unions were frequent, as is to be expected among a family living in an area that has been isolated for centuries (Moroni et al., 1972). As a direct consequence, the variety of family names contained in the genealogy reconstructed is small. The small variety, however, can be attributed only in part to the small size of the community. We were able to trace the descendancy of all relatives with the surname Deiana to a single ancestor, Mattia, but were unable to do so for the Todde family, despite the fact that four marriages involved one Dejana to another Todde. The data available would allow the assumption to be made that there is a tendency towards consanguineous marriages; however, it does not provide sufficient information to substantiate the assumption. An obvious example of consanguinity is the marriage of a centenarian female cousin to a first degree cousin, who is also a first degree cousin of Antonio Todde.

3.2. The family genealogy of Giovanni Efisio Emanuele Frau

The decision to include Giovanni Efisio Emanuele Frau's family genealogy was based on grounds similar to that for Antonio Todde. The former is the only other "full-fledged" supercentenarian.⁸ He, too, was included and his documentation validated first in AKeaA 1, then in AKeA 2. He was born in an Inland Municipality in the Province of Nuoro, where he died at the age of 112 years, 5 months and 20 days.

Civil status registers for the municipality of both Orroli and Tiana are complete for three series (births, marriages, and deaths) from the year 1866. They are well preserved, easily accessible, written in Italian, and easy to read. For life events prior to 1866, we used the digitalized records of the parish registers stored at the Sassari Regional Archive. These sources are accessible, written in Castilian until about 1820, then in Latin, and they do not have important gaps, although the older documents are occasionally damaged and quite difficult to read.

Parish records have been used also for the years after 1866 in order to increase the accurateness of the validation procedure.

As for the Todde family, information on Giovanni Efisio Emanuele Frau and his family was consistent with the various birth, death, and marriage certificates. A similar validation procedure and construction of the family biographies was also conducted.

The reconstruction of Frau's family history was successful despite a number of gaps, such as in the kinship network,⁹ owing to the fact that Orroli was fairly isolated and closed to the outside world (see Fig. 2).

⁸ In November 2003, about two months after the cut-off date of our sample (September 15), another supercentenarian celebrated his 110th birthday (he died before his next birthday).

⁹ For earlier periods, the correct identification of kinship in Orroli is complicated by the fact that at times in the early 1700s female offspring were often given their mother's surname.

Mr. Frau was born on December 29, 1890 in Orroli and died there on June 19, 2003. At that time, he was the oldest man in Europe. He worked as a farmer all his life.

His father was aged 45 when he was born, and his mother 32. Giovanni married Giuseppa Cavalleri in 1927 at the age of 36. His wife died in 1995, aged 89. They had eight children, and the last was born when Giovanni was 56. At the date of the survey, six of his children were still alive: of the other two, one died at the age of 3, and the other aged 68.

Various cases of longevity are found in his family. Of his seven siblings, a sister died aged 95 years, one brother aged 90, and another aged 84.

Some other longevity cases¹⁰ were found in the maternal branch of the family (five generations included). Giovanni's mother, Daniela Fortunata Orrù, born in 1859, died aged 84, and two aunts died at the ages of 82 and 79, respectively.

Giovanni's maternal great-grandparents were related: the parish marriage certificate contained a dispensation for "4th degree kindred", presumably cousins. It is because of this dispensation that kinship could be traced. Merely tracing the biographies of the ascendants of the individuals of this family did not reveal all of the kindred relationships existing.

Going back in time, a case of longevity was found in the Orrù's family: a maternal grand uncle, Gianuario Luigi, born on February 26, 1815, died at the ripe age of 88.

Interestingly, the other centenarian from Orroli included in the project is a relation of his: Elena Orrù, born in 1903. She died in May 2004 at the age of 101. Giovanni Frau and Elena Orrù's grandfathers were, in fact, brothers. Elena Orrù's is Antonio's daughter (she died aged 72), who is Gianuario's son.¹¹ When Elena, the second last of seven children, was born her father was 58 years old and 65 when the last son was born. Two of Elena's brothers died when they were over 80 years old: one aged 88 and the other aged 82.

Recently, the Municipality of Orroli published on its website a list of over 90 year-olds living in this region on March 1, 2004. Of a total of 33, 20 were women and 13 men. Four of the women, including Elena (aged 100), had the same surname, Orrù. Care should be taken, however, to make assumptions based on this as it is the most frequent surname among these generations. Another woman, Ottanza, born in 1912, is a first degree cousin of Giovanni Frau. Ottanza is the daughter of Battista, the niece of Daniela Fortunata Orrù, Giovanni Frau's mother. Daniela's brother, Tommaso, is Battista's father.

Not a single ascendant has been traced for the other Orrù, although the frequency of certain names at baptism would lead us to believe that they have a common ascendancy. Only a careful reconstruction of family trees, going back in time as far as possible, will reveal the existence of kinship, however remote.

Two pairs of siblings were also included in the list of 33 ninety-year olds in Orroli (two brothers, born in 1905 and 1911, and a sister and brother born in 1909 and 1911, respectively). Intriguingly, a grandparent of the two brothers is called Fortunata Orrù, but kinship with Giovanni Efisio has not been established. Perhaps other forms of kindred exist among the other over 90-year olds, something that only further research will reveal.

3.3. The family genealogies of Antonietta Sebastina Serra and Caterina Serra

The family history of the two cousins above is a unique example of the history of inter-family relationships between spouses.

The reconstruction of the family tree and the validation process for the two cousins, who were born in Padria and continued to live there, were identical to those performed for Todde. Documentation on both is well preserved and easily accessible.

Antonietta Sebastiana, who died at the age of 106, was the daughter of Caterina's mother's brother (she died aged 107 years). Antonietta Sebastiana's mother's died at childbirth at the age of 22, the father died aged 67 years. Caterina's mother died at an older age, at the age of 93 (the 1877 generation), and her father died aged 73. Caterina's parents had the same surname as they were second cousins, confirmed by a dispensation for 4th degree kinship from the bishop, contained in the parish marriage register.

The cousins' parents come from large families. The grandfather married a woman 28 years younger. He had 10 children, three of whom died at the ages of 93, 93, and 88, respectively. Caterina's mother, who died at the age of 93, was born when her father was as old as 65, and his wife was aged 37.

The grandfather and grandmother, common to both cousins, died at the age of 87 and 85 years, respectively. They both came from long-living families. The paternal grandfather of both cousins had a maternal grandmother who died when she was 88 and had the surname of the mother, although no clear kindred between the two was revealed. Longevity runs in the family branch shared by Antonietta Sebastiana and Caterina. The family history of Caterina contains another curiosity: Caterina was born when her father was already almost 70, she then married her cousin, the son of her maternal grandfather's nephew, who is the son of the grandfather's brother. Another peculiarity is that Caterina's paternal grandfather has the same surname as the rest of the family, although once again no

¹⁰ Particular instances of longevity were not identified along the paternal line, apart from two paternal great-grandparents: a great grandfather born in 1770 who died aged 77 and a great grandmother born around 1775 who died aged 75, as stated on the death certificate (but not corroborated). All other instances of longevity mentioned in this paragraph were validated. ¹¹ We focused only on Elena Orrù's direct ascendants. Antonio, for instance, is not Gianuario's only child. This analysis will be extended to include Antonio's other siblings and their descendants.

direct kindred between the two families was established. The only conclusion that can be drawn from this is that four generations of the ascendants of Caterina's family have the same surname.

Antonietta Sebastiana and Caterina's family genealogies, from the mid-1700s until today, only contains three surnames of three families, each of which has a history of long-living.

4. Discussion

The clustering of exceptional survival within families suggests a familial component affecting mortality differences, especially at extreme old ages, as in other centenarian genealogical studies (Robine and Allard, 1997). But of course not all familial effects are genetic. That an exceptional number of persons in the same family reach extreme old ages does not necessarily imply that genetic factors play a role in human longevity. Shared characteristics, such as life style and the environment, might also be involved.

An important aspect emerging from this centenarian study is that longevity was found among the ascendants of a particular branch of the family, and in most cases along the maternal line, as in other studies (i.e., Jalavisto, 1951; Abbott et al., 1974; Philippe, 1978; Bouquet-Appel and Jakobi, 1991) while, on the contrary, other authors (Bell, 1918; Wyshak, 1978) claim that the role of the male line is greater than that of the female line. The sex dependence of the inheritable component of longevity might suggest cytoplasmic (mitochondrial DNA) or sex chromosomes-linked inheritance (Cournil, 2001).

Our data are still provisional, should it apply to other validated cases, it would provide empirical evidence of a genetic component in longevity.

A more thorough examination of the data available may provide deeper insights into the role played by endogamy and consanguinity among Sardinians who enjoy exceptionally long lives.

The provisional results from the family histories considered in this study suggest the need for a long term analysis of certain communities, using all data available from the early 1800s, where there is a history of longevity. It should focus on the individual and family histories of all members of the community to corroborate the existence of longevity in the past and the presence of family clusters of longevity in the community or to find out whether longevity is transversal across families.

Answers to these questions would help to gain deeper insights into the role of genetic and environmental factors in long-living individuals and to verify the biodemographic existence of a longevity model common to sub-population groups.

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