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Lost in Transition – The Island of Susak (1951–2001)

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A B S T R A C T

The isolated population of the Island of Susak was thoroughly studied by a multidisciplinary team of the Croatian Academy of Sciences and Arts in early 1950's. Recently, a team of scientists revisited the island. This paper describes the main characteristics of the transition process during which a massive exodus occurred with 90% of the island's population migrating to New Jersey, USA. We summarise the differences in lifestyle, economy, social structure and sense of identity between the historic (1950's) and contemporary (2001) Susak population. We applied contemporary methods (analysis of microsatellite DNA polymorphisms) to investigate local myths about extreme levels of inbreeding and genetic homogeneity among the Susak islanders. Analysis of short-tandem-repeat (STR) loci showed that Susak displayed characteristics of a small homogeneous breeding isolate. The average heterozygosity was found to be low compared to outbred populations. The signature of a recent severe bottleneck could be detected. Analysis of 8 markers located on Xq13–21 in 71 individuals suggested extensive level of linkage disequilibrium (LD). A migrant study was designed to investigate the effects of large environmental changes (Susak vs. USA) and inbreeding (Susak vs. Croatian general population) on some biologically important quantitative traits, such as blood pressure and serum lipids. The results confirmed the positive correlation between inbreeding level and blood pressure that has been reported in the literature on several occasions. The last remnants of this traditional island community will soon be lost forever.

Key words: *island of Susak, Croatia, bottleneck effect, anthropology, migrant study, inbreeding, quantitative traits, quality of life, SF-36 questionnaire*

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Introduction

Susak is the most remote inhabited island in the northern Adriatic Sea. It is a small, low plateau of sand on a limestone base with a circumference of 3.75 km^2 (Figure 1). In the early 1950's, the Academy of Sciences and Arts (previously »Yugoslav« and currently »Croatian«) launched a multidisciplinary study of the island. This island was incorporated administratively into Croatia, its motherland, at the end of the World War II. The traditional name of the island is *Sansego,* derived from »*sampsion*«, a Greek name for the small blue flower abundant on the island. The vineyard growers and fishermen of this isolated island retained their traditional way of life, their habits and social structure and their dialect of the Croatian language. It was evident that with time the island and its people would be subjected to changes, and the Academy decided to study the community in order to learn more about it, and to obtain a baseline reference for possible follow-up. The results of those extensive studies were published in a monograph in 1957[1]. This book presented an in-depth analysis of the island's geographic and ecological distinctions, geologic and hydrologic features, socio-economic status and lifestyle, population dynamics, demography, schooling, crime, vineyard growing and fishing. Further anthropological studies were undertaken into characteristics of the archaic dialects used by the islanders, their lifestyle and clothing, folk songs, folk music, dwellings and architecture. Anthropological investigation collec-

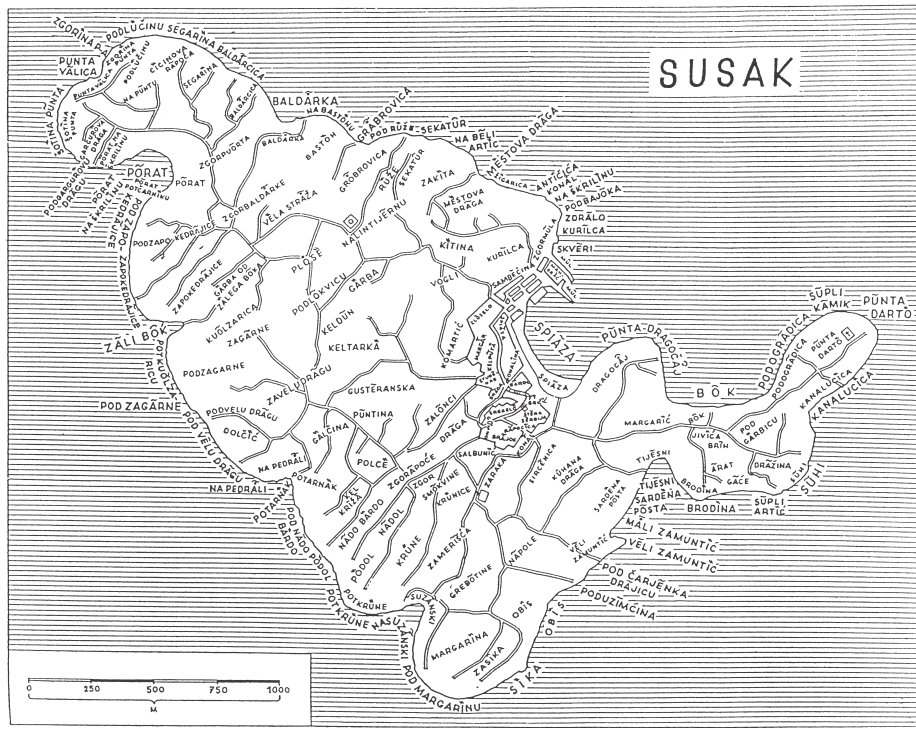

Fig. 1. The island of Susak.

ted data on child growth, adolescence and adult anthropometry, blood groups and genealogy structure. In the chapter on health and diseases, sanitary conditions, prevalent infectious diseases and other illnesses and disabilities among the islanders were described. However, in the period between 1951 and 2001, the island and its community went through deep transitional changes. The large majority of the population (about 90%) emigrated to the United States of America, mostly to Hoboken, New Jersey. This led to the traditional community quickly vanishing. Agriculture and fishery were abandoned, with newcomers developing tourism and turning the island into summer tourist resort.

The aim of this paper is to make an anthropological assessment of the differences within this extremely isolated community over the period 1951 to 2001. Some of the apparent characteristics of the island population of Susak population in 1951, such as high levels of population inbreeding, could now be assessed and quantified using modern methods based on the analysis of DNA polymorphisms. Furthermore, the unique »natural experiment« of genetically homogenous population living in the two very different environments of Susak island, Croatia and Hoboken, NJ, USA, provided the opportunity to assess the relative importance of genetic and environmental influences on several important biological traits, such as blood pressure and serum lipids, and on health-related quality of life.

Materials and Methods

Studies of changes during transition process (1951–2001)

The follow-up field work on the island of Susak was performed in 2001. This was done through several visits to the island by the team from the School of Public Health »Andrija Štampar« of the Medical School, University of Zagreb, Croatia and Croatian Institute of Public Health. The transition process on the island of Susak was characterised qualitatively, based on historic documents and interviews with the villagers from the demographic and socio-political perspective. Issues related to the lifestyle of the islanders, their economy, social structure and identity were compared in 1951 and 2001[1–7].

Studies of genetic variants (2001)

A sample of blood was collected into an EDTA tube (for DNA extraction) and frozen at –20 °C. Another blood sample was collected for biochemical analyses. Within one month, DNA extraction was performed at the University Clinical Hospital Centre in Zagreb, Croatia. The DNA was sent by overnight carrier to the Human Genetics Unit, Medical Research Council, UK, where genotyping of microsatellite markers was performed. Ten nanograms of DNA was amplified using fluorescent primer-pairs and the genetic linkage panel 11 (Applied Biosystems) for autosomal markers, or synthesized labeled primers (based on sequences from Genome Database GDB, http://www.gdb.org) for the X-linked markers.

Genotyping was analyzed using the Genotyper software (Applied Biosystems) following separation of the fluorescently tagged PCR products using an ABI3700 DNA sequencer. Allele frequencies for each microsatellite locus were estimated using Genepop software (http://wbiomed.curtin.edu.au/genepop/). Eighteen microsatellite markers (D7S484, D8S264, D8S260, D7S517, D8S178, D7S246, D8S549, D8S258, D7S669, D8S272, D7S502, D7S630, D7S510, D7S640, D7S513, D8S514, D7S516, D8S1771), at least 5cM apart, were used to test for departure from random-mating. Nei's average heterozygosity, or gene diversity,

was calculated as one minus the sum of the squared allele frequencies[8]. Only loci which have been previously investigated in other populations were used for gene diversity calculations.

For analysis of the extent of linkage disequilibrium (LD) in the contemporary population, ten X-linked microsatellites were genotyped. Eight of them were Xq 13–21 linked. These encompassed six of the markers described by Laan and Paabo[9]: DXS983, DXS8092, DXS8037, DXS1225, DXS8082, and DXS995. Those six markers were complemented with additional two interspaced microsatellite markers: DXS1165 and DXS56. The complete Xq13–21 region with these eight markers spans 3.36 cM[10]. To investigate LD patterns at genomic distances that are an order of magnitude greater, two more markers (DXS8085 and DXS8014) were genotyped. They are located at Xp21 and situated, respectively, 18 cM and 23.68 cM away from the most proximal Xq13 marker (DXS983)[9]. Female haplotypes were inferred using a Bayesian statistical method implemented in PHASE (v2.0)[11]. The algorithm was run 5 times and the run with the best average goodness-of-fit kept. At each locus, only those genotypes for which phase certainty was >80% were further analyzed. Such inferred female haplotypes and the males haplotypes were used to calculate a pairwise measure of LD, D' corrected by permutations (1000 replicates) using the software miLD developed by Aultchenko et al.[12].

Migrant studies of quantitative traits (2001)

Given the unique opportunity that this population in transition presented, we studied the effects of inbreeding and environmental change on the value of a number of biologically important quantitative traits (QT's) which are established as major risk factors for health. During the visits to the island, we measured systolic and diastolic blood pressure, total and HDL cholesterol and triglycerides among the native islanders and visitors to the island who had lived in the USA longer than 20 years. For each person, we selected 4 controls from the general Croatian population matched by sex and age. Controls were chosen from a large database of 10,000 persons from the general population of Croatia who had attended the biochemistry laboratory at the University Hospital Center »Zagreb« in Zagreb, Croatia[13].

We performed two comparisons between the cases and controls. Firstly, we compared the mean values among native Susak population and matched controls from the general population of Croatia, which should reflect the effect of inbreeding on changes in mean QT values. Secondly, we compared mean values among the group of Susak emigrants with controls from the general population of Croatia (Table 1), which should reflect the combined effect of environmental change and inbreeding on measured QT's. Differences in means were assessed using the Student's t-test. We did not compare the mean values between Susak natives and emigrants to USA, as the number of observations was too low to achieve sufficient power for meaningful comparison.

Studies of health-related quality of life (2001)

Health-related quality of life was measured using the SF-36 questionnaire, a well-known and widely used instrument of self-assessed health status[14]. It measures a number of dimensions of health status, such as physical, psychological and social functioning, role fulfillment within a community and perception of one's own health. Large quantities of data were collected, but in this paper we only presented mean scores on the eight main dimensions of health status. Mean

TABLE 1
DESIGN OF THE MIGRANT STUDY OF THE EFFECTS OF INBREEDING AND ENVIRONMENTAL CHANGE ON SIX QUANTITATIVE TRAITS (SYSTOLIC AND DIASTOLIC BLOOD PRESSURE, BODY MASS INDEX, TOTAL CHOLESTEROL, TRIGLYCERIDES AND HDL CHOLESTEROL)

Step 1 (visit 1):	Measured quantitative traits (QT) – blood pressure, height and weight – in 72 persons (32 Susak natives and 40 visiting emigrants to Hoboken, NJ, USA)
Step 2 (visit 2):	Blood collected for genotyping from 72 persons – analysis of 18 autosomal and 10 X-chromosome polymorphic microsatellite markers
Step 3 (visit 3):	Blood collected for biochemical analyses (total cholesterol, triglycerides and HDL cholesterol) in a limited sample: 23/32 natives and 12/40 emigrants
Step 4:	Four sex and age-matched controls were found for each examinee from the large database of 10,000 persons from the general population of Croatia, analyzed by the same laboratory at the University Hospital »Zagreb« in Zagreb, Croatia
Step 5:	Comparison that should reveal effects of inbreeding: mean QT values in native Susak population were compared to mean QT values in matched controls from the general population of Croatia
Step 6:	Comparison that should reveal combined effects of environmental change and inbreeding: mean QT values in emigrant Susak population were compared to mean QT values in matched controls from the general population of Croatia

scores represent the averages of achieved scores in each dimension (on a scale from 0 to 100) with 0 being the worst possible health, and 100 being the best. The dimensions which were studied were physical functioning (PF), role physical (RP), bodily pain (BP), general health (GH), vitality/energy (VT), social functioning (SF), role emotional (RE) and mental health (MH). Change in health (HT) which is a separate item characterising the dynamic aspect of health was also investigated. The results were compared with those already reported for a number of countries, including the general population of the Republic of Croatia[15,16].

Results

Characterizing the transition process (1951–2001)

For about a quarter of the 20th century (1919–1945), the island of Susak was under Italian rule. During that time, Italian was official language in schools and all other institutions. The island's name was »*Sansego*«, and islanders were called »*Sansegotis*«. After the World War II, the island was reintegrated into the mother country, the Republic of Croatia (part of the Yugoslav federation at the time). The mother tongue was reinstated in schools, institutions and public life. The economy also changed from capitalism to socialism. The despised fascist dictatorship was replaced by a single-party political system with a socialist ideology. Those changes were extremely abrupt and far too rapid for a small, traditional insular community to accommodate.

The main problem during the reintegration was that traditional dialect spoken on Susak was now hardly understood by mainland Croats, which left the »*Sansegotis*« feeling very isolated. »We feel as we are embarked on a sailboat«, declared

an elderly lady. It seemed that many, especially the young, were looking for a way to »disembark«[1]. There was an agricultural overpopulation and poor economic prospects for their relatively primitive wine growing, fishing and household management methods which were all based on intensive manual labour (Figure 2). There was also little hope for the development of modern industrial production, which could provide employment for all.

The community was very close and compact at the time – about 1,500 inhabitants shared only 8 different surnames. Related by blood, common faith and unique habitat, their solidarity was high and they were reluctant to leave the place to which they all shared strong feelings of belonging. However, economic needs prevailed among the young, who found a compromise in emigration to New Jersey, USA. This was a place where several of their relatives had already established themselves, but also retained close relations with the community remaining on Susak.

The transition on Susak was characterised by a massive emigration of the islanders to the USA between 1950 and 1980 (Table 2). By the end of the 1950's, there were few grapes to be harvested and the winery ended production and bottling. The level of fishing activity was insufficient to maintain the fish cannery.

Fig. 2. An illustration of the hardship experienced by island of Susak inhabitants during the mid-20th century: small girl washing her family's clothes by hands (Photograph by B. Cvjetanović, 1957).

TABLE 2
RECORD ON EMIGRATION FROM SUSAK TO THE UNITED STATES OF AMERICA (1950–2000)

Year	Population		Notes on emigration and its effects
	On Susak	Emigrated	
1950	1,551	28	During this decade, authorities hampered the emigration but did not effectively prevent it. Total population decreased by 30%, but the traditional way of life persisted. However, exodus of youth depleted the available workforce.
1951	1,512	28	
1952	1,473	39	
1953	1,438	34	
1954	1,384	35	
1955	1,322	38	
1956	1,249	46	
1957	1,171	52	
1958	1,114	37	
1959	1,049	46	

1960	982	40	From 1963 emigration was permitted. more than 60% of the remaining population emigrated during this decade. Shortage of workforce led to collapse of organised wine production and fishery activities.
1961	927	35	
1962	864	45	
1963	807	44	
1964	634	39	
1965	544	35	
1966	519	51	
1967	453	46	
1968	408	52	
1969	367	49	
1970	347	52	Additional 30% of remaining population migrated to United States of America. The rate of emigration slowed down in this decade, however, as most of those who intended to leave the island succeeded. The emigration ended rather abruptly in 1980.
1971	327	47	
1972	321	47	
1973	295	35	
1974	289	47	
1975	258	44	
1976	251	44	
1977	256	31	
1978	249	34	
1979	238	42	
1980	227	–	During this decade, after all the young and fit inhabitants migrated, the remaining population of the island (mainly elderly and disabled) has stabilised at about 200.
1981	220	–	
1982	209	–	
1983	199	–	
1984	197	–	
1985	192	–	
1986	187	–	
1987	190	–	
1988	186	–	
1989	190	2	
1990	188	–	Later in this decade, after the end of the War in Croatia, several people returned to the island from the United States. However, this had very little effect on the general result of the exodus.
1991	189	6	
1992	180	–	
1993	179	–	
1994	165	–	
1995	208	–	
1996	219	–	
1997	209	7	
1998	222	–	
1999	203	6	
2000	205	–	

The terraces of the vineyards were collapsing and invasion of reeds and bushes changed the landscape. Many homes were deserted and were falling into ruins, while others were sold to tourists, mainly from Slovenia and Austria, looking a secondary home for rest and retirement or for a quiet retreat. Others were caterers from the Croatian mainland, who were attracted by the economic potential of tourism on the island.

This trend increased the interest of the expatriates from the USA into their patrimony on Susak. Some bought back the houses of their parents, restoring their native homes with modern amenities and changing them into villas and even small palaces. The appearance of the village was changing. Through donations, the expatriates supported the restoration of the church and the construction of the community centre with the post office, outpatient clinic, retirement home and the premises for the »Club of expatriates«. The streets were paved, water cisterns built, and it became customary for the expatriates to come to Susak for annual gatherings in July or August. Some 200–300 expatriates would come and actively participate in the religious events, the folkloric dances with colourfully dressed girls (Figure 3), traditional singing and music. By this means the expatriates were proudly reviving the traditions of their ancestors while the tourists enjoyed these festivities.

At the end of the year 2000, there were 67 families with 205 members declared as residents of Susak. Among them, only 153 were born on the island, and 67 were newcomers. The newcomers introduced some »continental« (and even international) spirit. Croatian radio and television stimulated islanders to learn the literary Croatian language. Only the elderly continue to use their traditional dialect, which is gradually disappearing from daily use. Newcomers brought economic

Fig. 3. Traditional clothing specific of the island of Susak (Photograph by B. Cvjetanović, 1957).

revival through tourism, prompting some of the native islanders to revive their old vineyards for sale and domestic use. Some of them lived on the island all year round, adopting local ways of life and living in harmony with the local population, while others, motivated primarily by seasonal economic interest, were not as welcome. Elderly expatriates who decided to retire on Susak were often disappointed with the changes they found, in spite of many evident improvements such as electricity and telephone. The island they found had little movement and few activities with hardly a fishing boat in the port, many uninhabited houses, collapsed wine terraces, ruins of winery and fish cannery and rare elderly passers-by. The island was in deep transition.

The islanders (1951 vs. 2001)

In 1950, there were 1,551 inhabitants on Susak. In view of the small surface area of the island, the population density (403 per km^2) was the highest in the entire federation of Yugoslavia. The only link with the outside world was a small

wooden boat that sailed to the nearby island on working days, weather permitting. There were no public telephones, and radio connections served only the lighthouse overseen by the Yugoslav Army. There were no means of animal or motor transportation and everything had to be carried by people themselves. Diesel generator provided electricity until 10 p.m. and then the only light on the island was that of the lighthouse. All the inhabitants were familiar with each other and there were no secrets, as gossiping would take place daily while fetching the water at the community well. Each member of the community was thus obliged to behave properly in order not to be criticised and possibly rejected by the community. The cohesion of the community and harmonious interrelations among its members was treasured by all, and the islanders were very emotional. In their dialect, they had means to express their emotions by using an augmentative, diminutive or pejorative by adding an appropriate suffix in each case. This openness of expression of views and feelings was valued, as it strengthened the mutual trust and the cohesion of the community.

As to their ancestry, the islanders' recollection does not go farther than the inscriptions on the tombstones at the local cemetery. In 1701, it was reported that there were some 300 people living on the island. Two centuries later, there were 1,381 souls. The largest population (1,656 persons) was reached in 1936, following by the emigration and decline. In the 1950's, the community was living in their traditional, peculiar way of life. People were occupied during the day with vineyards and domestic work, while in the evenings many of them would go fishing. On Sundays people would go to church in the morning. In the afternoons they would gather on the main square for dancing and gossiping. As many individuals shared same names and surnames, nicknames were given to individuals. Nicknaming thus became customary on the island and it was also applied to visitors. Usually, this would occur at the time the boat docked at the pier and the passengers were disembarking. The crowd of islanders that were there to greet every arrival would briefly discuss and agree on a nickname that they felt best suited the person concerned. Even the heads of the state were not spared of this custom. In 1953, Yugoslav president Tito was named »*Galebina*« (a large seagull), in 1994 the first Croatian president Tuđman was nicknamed »*Hinić*« (a pretending person), while in 2000 the second Croatian president Mesić acquired the nickname »*Barbučić*« (a small beard). The islanders were critical and straightforward with each other, and they treated visitors in the same way.

Economy (1951 vs. 2001)

Natural resources were the fertile sandy land and the fish rich sea. Part of the agricultural products and fish were for the domestic use and the other part for the market. This was a mixed, subsistence and marked economy. The main product for the market was wine, while fish was mainly sold to the local fish cannery that employed local manpower. Nearly all land available on the small island was turned into grape growing terraces. The economy was entirely relying on those two products, making it highly vulnerable to fall in wine prices or decreasing quantity of fish in the surrounding waters. With overpopulation, many islanders started to leave for the New Jersey, USA. Accustomed to the hard work at home, they found their ways in the new homeland and began to take part in industry and trade. Their income supported their families left behind on Susak. In 1954, the Ministry of Finances estimated that the expatriates contributed some 20% to the total income of the

islanders. The wine generated 55% and fishing another 15% of the earnings. It did not take long before the islanders realised that their relatives in the USA were much better off. The young people were attracted to the opportunity, especially with the assistance offered by the association of expatriates, which fuelled the emigration.

In 2001, the population consisted mainly of elderly persons who were dependent on support from their expatriate relatives. Very few were still fishing or producing wine, mainly for personal use. Some were also leasing rooms to tourists, while those who returned from abroad were receiving the retirement rights. The newcomers, however, were primarily engaged in tourist trade and catering. Between 1,000 and 1,500 overnight accommodations was registered annually in the last few years, with tourism being the only economic potential of the island for the foreseeable future. The great majority of tourists currently spend only one night on the island, and about 75% of them are foreigners. The key to expanding the current capacity of about 200 beds would be a water supply brought from the mainland undersea via nearby islands.

Social structure (1951 vs. 2001)

The social structure of the community has developed through history. The first owners of the island were religious authorities. The civilian society developed gradually and the islanders became the owners of the land they harvested. The attachment to the Catholic religion remained, and the church continued to play an important role in the community. Governmental authorities replaced the religious ones in economic, legal, educational and administrative matters, but they could never establish a deep control over the traditional social structures on this small and remote island. The community continued to obey their own unwritten traditional rules and the decisions from informal gatherings and consultations of seniors. The isolation and interdependence of people, who were much like a crew on a sailing boat, imposed an obligation to show solidarity towards the others. Disobeying the principle of solidarity would mean excommunication, with humble seeking of forgiveness or leaving the island being the only remaining options. However, the community was far from being vindictive. On the contrary – it was rather permissive and tolerant of human weaknesses, but not of the violation of solidarity. Crimes on the island were rare, as people adhered to traditional values and abstained from unacceptable behaviour.

The traditional community on the island was by emigration reduced to a small group of elderly who represented a minority in the population. However, their importance in the social life in the newly emerging community of natives and newcomers was great because they possessed practically all land, the majority of houses and other resources. They had strong financial and moral support from their relatives in the USA, who provided funds for the communal institutions. The newcomers contributed very little to these developments, if anything at all. In 2001, one of the natives was village headman, while the others were running local communal institutions. The newcomers, however, were handling local commerce and tourism. They brought their experience from the mainland and shared it with the islanders. The authorities supported both the natives (to prevent emigration) and the newcomers (who were boosting the economy by developing tourism). There was a clear interest of the authorities to sustain the development of an active, economically sustainable and socially stable community of islanders and newcomers emerging on Susak.

The expatriates in the USA retained their insular cohesion. They organised gatherings, societies and clubs to maintain their insular traditions and solidarity. The »St. Nicholas Society of Sansego« (St. Nicholas was the patron saint of Susak) was established with this in mind in 1948. It included the folkloric group and the soccer club and was embracing practically all of 2,647 expatriates and their family members in New Jersey. By their number and economic resources, they continued to play an important role in social life on Susak in spite of the geographic distance. The solidarity and the cohesion of the islanders who live on Susak and those in New Jersey could hardly be matched by any similar example in Croatia.

Identity (1951 vs. 2001)

The identity of the people of Susak was recognised fully by the islanders and also by the other (outside) people. Islanders considered themselves different from the others by their dialect, habits, folklore costumes and the way of life on their unusual island covered with vineyards. While the islanders perceived their identity as a sign of their superiority to the others, their neighbours considered them inferior because of their consanguinity and related stereotypes and also due to rather primitive ways of their labour in the fields. The islanders had multiple identities: besides their collective identity, they had a lineage identity (i.e. belonging to one of the eight large traditional families with common family names), and their individual identity[17,18]. However, the most important was their collective identity as the Susak islanders. This was, however, not valued in a similar way in their new homeland, the USA. There, they faced a society focused on individualism and competition. The expatriates suddenly faced conflict between their individual and collective identities. Their individual, rather than »lineage« identity, gradually became the basis for their »new« American identity.

The emigrants to the USA were exposed to the process of assimilation as soon as they were involved in economic and social activities. Mixed marriages began to replace inbreeding. Children attended American schools and the knowledge of the mother tongue was not readily transmitted to the young, let alone the written word. Traditions were maintained only through gatherings of the »St. Nicholas Society of Sansego« and visits to Susak. The islanders soon faced an »identity crisis«, much like any emigrants who leave small homogenous communities for a large, multicultural state like the USA. However, the islanders apparently did not lack adaptability – they emphasised their attachment to religion, which is also highly valued in the USA, and used the freedom of expressing their traditions (folk music and dance) through the activities of the »St. Nicholas Society of Sansego«.

Genetic studies (2001)

In the whole of Croatia, the island of Susak represents a paradigm for a place of the most extreme isolation and inbreeding. Outsiders perceive the islanders as highly consanguineous. One of the aims of this paper was to investigate the accuracy of those myths using contemporary methods (analysis of microsatellite DNA polymorphisms). In terms of allelic and gene diversity, most of the loci for which genotyping information existed in other populations displayed reduced allele diversity (a reduced number of alleles despite large sample size) and gene diversity consistent with the existence of a reduced gene pool (Tables 3 and 4). Analysis of 18 autosomal markers in 70 individuals was consistent with a population undergoing random mating suggesting that the population was homogeneous.

After going through a bottleneck, a population will have a transient excess of heterozygosity compared to that expected for the allele number, as the number of alleles at a locus is reduced faster than gene diversity (rare alleles are more affected by drift than more common ones). The equilibrium gene diversity (Heq) was computed from the observed number of alleles and under the assumption of a constant-size (equilibrium) population using the program Bottleneck[19]. There was a significant excess of gene diversity in Susak compared to the level expected at equilibrium (Wilcoxon test one tailed test, p=0.049) indicating a clear reduction of effective population size (Table 5).

TABLE 3
COMPARISON OF SUSAK GENE DIVERSITY BASED ON AUTOSOMAL MICROSATELLITE MARKERS WITH CORRESPONDING PUBLISHED GENE DIVERSITIES FROM ISOLATES OR OUTBRED POPULATIONS (DATA FOR YAKUT, DANES AND JAPANESE POPULATIONS FROM http://info.med.yale.edu/genetics/kkidd/contents.html)

Marker	Diversity (No of alleles)			
	Susak 2N=142	Yakut 2N=102	Danes 2N=118	Japanese 2N=100
D7S484	0.593 (6)	0.819 (8)	0.684 (6)	0.781 (7)
D8S260	0.788 (8)	0.829 (9)	0.803 (9)	0.807 (9)
D7S517	0.786 (9)	0.801 (7)	0.833 (10)	0.763 (7)
D8S258	0.567 (4)	0.676 (4)	0.746 (7)	0.657 (5)
D7S669	0.781 (8)	0.778 (8)	0.771 (10)	0.798 (9)
D8S272	0.471 (8)	0.807 (9)	0.757 (10)	0.819 (8)
D7S510	0.734 (6)	0.668 (7)	0.778 (10)	0.802 (8)
D7S640	0.836 (15)	0.869 (16)	0.872 (17)	0.889 (20)
D7S513	0.814 (13)	0.815 (13)	0.836 (14)	0.899 (15)
D8S514	0.698 (6)	0.762 (6)	0.656 (7)	0.712 (8)
D7S516	0.779 (5)	0.697 (7)	0.704 (6)	0.695 (6)

TABLE 4
COMPARISON OF SUSAK GENE DIVERSITY BASED ON X-LINKED MICROSATELLITE MARKERS WITH CORRESPONDING PUBLISHED GENE DIVERSITIES FROM ISOLATES OR OUTBRED POPULATIONS

Marker	Diversity (No of alleles)				
	Susak N=118	Saami[9] N=54	Khoton[24] N=40	Swedes[9] N=41	Eur. Amer.[24] N=49
DXS983	0.64 (5)	0.58 (4)	0.66 (6)	0.62 (6)	0.76 (8)
DXS8037	0.63 (6)	0.50 (5)	0.53 (3)	0.78 (7)	0.68 (6)
DXS8092	0.82 (9)	0.83 (8)	0.88 (11)	0.89 (11)	0.81 (10)
DXS1225	0.73 (7)	0.76 (6)	0.86 (9)	0.81 (8)	0.77 (10)
DXS8082	0.65 (8)	0.79 (6)	0.84 (7)	0.76 (7)	0.78 (8)
DXS995	0.58 (5)	0.44 (3)	0.55 (4)	0.56 (5)	0.59 (6)

TABLE 5
TEST FOR THE PRESENCE OF A RECENT GENETIC BOTTLENECK. LISTED ARE THE OBSERVED (Ho) AND EXPECTED EQUILIBRIUM (Heq) HETEROZYGOSITIES GIVEN THE OBSERVED NUMBER OF ALLELES AT EACH LOCI (ko). Heq EXPECTED UNDER MUTATION/DRIFT EQUILIBRIUM WAS COMPUTED BY THE PROGRAM BOTTLENECK (http://www.ensam.inra.fr/URLB) USING A MIXED MUTATION MODEL FOR THE MICROSATELLITE MARKERS (70% STEPWISE MUTATION MODEL). DH/sd IS THE STANDARDIZED DIFFERENCE BETWEEN OBSERVED AND EXPECTED HETEROZYGOSITIES AND p IS THE PROBABILITY OF THE OBSERVED HETEROZYGOSITY AT A GIVEN LOCUS

Locus	N	ko	Ho	Heq	SDeq	DH/sd	p
D7S484	134	6	0.593	0.647	0.106	–0.502	0.2460
D8S264	140	7	0.809	0.691	0.094	1.261	0.0420
D8S260	138	8	0.788	0.725	0.084	0.757	0.2360
D7S517	136	9	0.786	0.759	0.071	0.382	0.4340
D8S178	134	5	0.576	0.577	0.135	–0.007	0.4020
D7S246	134	7	0.780	0.693	0.097	0.904	0.1670
D8S549	136	4	0.507	0.500	0.0148	0.046	0.4200
D8S258	138	4	0.567	0.497	0.152	0.459	0.3880
D7S669	132	8	0.781	0.727	0.081	0.655	0.2900
D7S502	130	8	0.840	0.729	0.082	1.344	0.0160
D7S630	134	8	0.795	0.728	0.084	0.795	0.2120
D7S510	126	6	0.734	0.645	0.110	0.812	0.1990
D7S640	138	15	0.836	0.864	0.035	–0.799	0.1730
D7S513	134	13	0.814	0.840	0.042	–0.631	0.2200
D8S514	132	6	0.698	0.637	0.112	0.549	0.3710
D7S516	132	5	0.779	0.581	0.127	1.561	0.0060
D8S1771	128	9	0.659	0.763	0.067	–1.544	0.0800

Another genetic investigation performed on the islanders was that of the extent of linkage disequilibrium (LD). This was done because a number of demographic factors in this population (e.g. relatively recent population history, large bottleneck, high inbreeding, low genetic diversity, genetic drift) were suggestive of, possibly, a very large extent of LD in the genome. Figure 4 shows that significant levels of LD were observed for all 28 possible pairs of markers spanning 3.36 cM in region Xq13–21. This indicates a high level of LD in the population. In comparison to other isolated populations reported in the literature this level is amongst the highest reported[9,11]. The other two markers (DXS8085 and DXS8014) that were respectively 18 cM and nearly 24 cM away from the Xq13–21 region failed to show significant LD with any of the other 8 markers. This finding was expected and it ruled out recent admixture as an explanation for the significance of LD observed.

Migrant study of the effects on quantitative traits (2001)

Table 6 shows the results of the migrant study, in which samples from Susak natives and Susak emigrants were compared with controls from the general Croatian population. Mean values of body mass index, systolic and diastolic blood pressure, total and HDL cholesterol and triglycerides were compared. Native inhabitants of Susak did not differ significantly in mean body mass index from the

Susak	1	2	3	4	5	6	7	8	9	10
DX–	S8014	S8085	S983	S1165	S8092	S8037	S56	S1225	S8082	S995
1										Dadj=D'-D'stoch
2	0,173									0.15<Dadj<0.25
3	0,018	–0,044								0.25<Dadj<0.5
4	–0,088	0,081	0,395							Dadj>0.5
5	0,090	0,066	0,277	0,527						
6	0,033	0,059	0,326	0,519	0,573					d(1,3)=23cM
7	0,190	0,067	0,206	0,431	0,475	0,554				d(2,3)=18cM
8	0,158	0,078	0,461	0,482	0,373	0,437	0,452			d(3,10)=3.36cM
9	–0,029	0,096	0,341	0,596	0,477	0,600	0,503	0,607		d(5,10)=0.85cM
10	–0,027	0,130	0,195	0,278	0,504	0,248	0,245	0,373	0,436	
	Xp 21				Xq 13–21					

Fig. 4. Linkage disequilibrium between 10 marker pairs on X chromosome.

general Croatian population, but the emigrants to USA had a significantly greater mean value. This is consistent with the suggestion that inbreeding did not affect body mass index significantly or has a much lesser effect than environmental factors in the USA. Susak islanders, living in both Susak and the USA, had significantly higher systolic (and, to a lesser extent, diastolic) blood pressure consistent with our previous reports of an effects of inbreeding on blood pressure[20–23]. Susak islanders had significantly higher HDL cholesterol and somewhat lower total cholesterol and serum triglycerides than the general Croatian population consistent with a favorable blood lipid profile. However, the sample sizes are small and these preliminary observations need to be taken with caution.

Health-related quality of life (SF-36) study (2001)

Table 7 shows averages of achieved scores (on a scale from 0 to 100) in each dimension of the SF-36 health status questionnaire. There were very few differences between Susak islanders and the general Croatian population. The main difference was a greater level of vitality/energy and a greater effect of emotional problems on ability to function in a community among the islanders. In comparison to other populations of the developed world, it is apparent that health-related quality of life is considerably poorer in Susak and Croatia than in developed countries for all eight dimensions[15,16].

Discussion

The transition that the island of Susak and its community are undergoing is complex and has many facets. The traditional insular, interdependent community of wine growers and fishermen on Susak has been reduced to a small number of elderly and disabled inhabitants who have been left on the island, while the rest have emigrated to the USA. The latter represents a community that continues to prosper in the new environment, adapting themselves to the multicultural and multiethnic American society. In order to meet the challenges of their new surroundings they have had to

TABLE 6
RESULTS OF THE MIGRANT STUDY, WHERE SAMPLES OF SUSAK NATIVES AND SUSAK EMIGRANTS WERE COMPARED WITH CONTROLS FROM GENERAL CROATIAN POPULATION. THE MEAN VALUES IN BODY MASS INDEX, SYSTOLIC AND DIASTOLIC BLOOD PRESSURE, TOTAL CHOLESTEROL, TRIGLYCERIDES AND HDL CHOLESTEROL WERE COMPARED

QT	Population	N	X	SD	p
BMI	Susak	30	25.88	4.06	ns
	Controls (S)	120	25.96	4.22	
BMI	Emigrants	38	28.01	3.49	< 0.05
	Controls (E)	152	26.48	4.12	
Systolic BP	Susak	32	160.27	31.84	< 0.05
	Controls (S)	128	143.12	28.40	
Systolic BP	Emigrants	40	155.75	26.07	< 0.05
	Controls (E)	160	146.02	24.96	
Diastolic BP	Susak	31	87.45	13.06	ns
	Controls (S)	124	82.95	11.03	
Diastolic BP	Emigrants	40	87.55	13.41	ns
	Controls (E)	160	85.32	13.34	
Total cholesterol	Susak	23	5.52	1.44	ns
	Controls (S)	92	5.68	1.39	
Total cholesterol	Emigrants	11	5.42	1.15	ns
	Controls (E)	44	5.73	1.22	
Triglycerides	Susak	23	1.31	0.50	ns
	Controls (S)	92	1.39	0.81	
Triglycerides	Emigrants	11	1.35	0.45	ns
	Controls (E)	44	1.49	1.12	
HDL cholesterol	Susak	22	1.67	0.66	< 0.05
	Controls (S)	88	1.30	0.45	
HDL cholesterol	Emigrants	12	1.55	0.59	< 0.05
	Controls (E)	48	1.19	0.41	

transform their identity based on a collective one (prized on Susak), to the individual identity which matters more in the USA. They are gradually assimilating into American society, while their kinsmen on the native island are mixing with the newcomers in a newly emerging community oriented to tourism. Transition is a process that continues among the members of the former traditional community, both among those left on Susak and those who emigrated to New Jersey. While the traditional community on Susak is vanishing, the expatriates to the USA are nostalgically nursing the traditional dances and music and restoring the paternal homes on Susak before being assimilated into American society.

The native community of Susak is vanishing from the island. The community of expatriate islanders presently centred in New Jersey is increasing in number and is gradually spreading throughout the USA and, in a few cases, to other countries. With the involvement in the economic and social structures of the receiving countries and mixed marriages,

TABLE 7
AVERAGES OF ACHIEVED SCORES IN EACH DIMENSION INCORPORATED INTO THE HEALTH STATUS (ON A SCALE FROM 0 TO 100) BASED ON SF-36 QUESTIONNAIRE (SOURCES FOR OTHER POPULATIONS: WARE ET AL.[15]; AUSTRALIAN BUREAU OF STATISTICS[16]; AND G.V., PERSONAL COMMUNICATION)

Scale	Susak	Croatia	U.S.A.	Spain	Sweden	Italy	Holland	Australia
Physical functioning (PF)	69.9	69.9	84.52	56.5	74.5	84.5	83.0	82.0
Role physical (RP)	63.2	63.0	81.20	66.5	71.8	78.2	76.4	78.5
Bodily pain (BP)	69.1	64.5	75.49	71.2	69.5	73.7	74.9	75.7
General health (GH)	55.3	53.4	72.21	42.2	69.8	65.2	70.7	71.9
Vitality/energy (VT)	60.4	51.9	61.05	58.3	97.9	61.9	68.6	61.9
Social functioning (SF)	72.6	73.0	83.60	82.4	86.8	77.4	84.0	86.9
Role emotional (RE)	65.8	72.4	81.29	75.3	75.6	76.2	82.3	84.6
Mental health (MH)	63.9	61.7	74.84	71.7	77.4	66.6	78.6	78.3
Health trend (HT)	49.3	44.8	–	–	–	–	–	–
Total	63.4	–	–	–	–	–	–	–

they will be assimilated into the society of their new homeland. However, a few families have returned from the USA to Susak. Some are trying to apply the knowledge and the resources gained abroad to revive the economy and make their living on the island, while others just want to renew their ancestral homes and use them as a peaceful retreat. The qualitative study among expatriates visiting Susak in the year 2001 revealed that the majority did (or wanted to) repair or rebuild the home of their ancestors for vacationing and possibly for the retirement, but not to return to permanently reside on Susak. Deep links between the expatriates and their native island will, inevitably, gradually fade away. However, the paternal home on the charming island will continue to attract the expatriates to visit Susak. For some, the ancestral traditions may still continue to be a part of their multiple identities. The newcomers will, together with the natives left on Susak, finally form a new community, probably less interdependent as they will not be so isolated and will be earning their living mostly from tourism rather than from heavy manual labour in vineyards. The island might again be covered by forest like in ancient times. The village will turn perhaps into a tourist resort flourishing in summer and being more or less deserted in winter. Some of the newcomers or natives may, for the sake of tourism or for their own amusement, revive the traditional folklore music and dances. Be as it may, the island will probably stay, but its traditional community will be gone.

This study used current scientific methods to describe this unique population of Susak in both a Croatian and an international context. The analyses of gene diversity and linkage disequilibrium based on genomic polymorphisms confirmed that the level of genetic isolation in the population is extremely high. Further genetic analysis will be required to estimate the real level of inbreeding in this population. Studies of linkage disequilibrium showed long regions in LD based on studies of microsatellite markers, with allele associations spanning distances of at least 3.3 cM. An analysis of health-related

quality of life using the SF-36 questionnaire showed that a summary measure of »physical health« (incorporating the 4 dimensions of PF, RP, BP and GH) was nearly identical to the general Croatian population. The summary measure of »mental health« (incorporating the 4 dimensions of RE, SF, MH and VT) revealed that the islanders have greater levels of vitality/energy, but their ability to function in a community is more affected by emotional problems.

The anthropological studies had an effect on the community. The multidisciplinary study carried out in the early 1950's gave a detailed picture of the island and its population, their way of life, language, habits, costumes, folklore and an account of the hard work of the islanders in the vineyards and their efforts and skills when fishing and sailing the seas. The published monograph, a voluminous book furnished with many photographs and maps, was acquired by almost all families. It was treasured and incorporated into their collective identity. The qualitative study of the expatriates revealed that about half of the families possessed it, and all of them wanted to have a video-copy of a film on the life on Susak in the early fifties. Their attachment to the native island and their perceptions about their origins and identity were greatly strengthened by this documentary book and the film. They became more aware of their unique historical origins and of the fact that it is rare to own such a detailed documentation of their ancestors, their community's characteristics and the evidence of their particular identity. It was noted that the traditional way in which the women combed their hair by pulling it tightly into a knot caused baldness (»*Alopecia gradus*«). They were advised on the harmful effects of such practices, which prevented further occurrence of this condition. The book and the documentary video will certainly help the survival of memories of this traditional community of fishermen and wine producers. It will extend for some time the cohesion of the expatriate community before their final integration into American society. The documents are likely to be treasured by the descendants of the ancient inhabitants of Susak into the distant future.

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IZGUBLJEN U TRANZICIJI – OTOK SUSAK (1951–2001)

S A Ž E T A K

Izoliranu populaciju otoka Suska temeljito je istražio multidisciplinarni tim Hrvatske akademije znanosti i umjetnosti (HAZU) u ranim pedesetim godinama 20. stoljeća. Ekipa znanstvenika je nedavno posjetila otok. Ovaj rad opisuje osnovne značajke procesa tranzicije otoka, tijekom kojeg se zbilo masovno iseljavanje i 90% otočnog stanovništva emigriralo je u New Jersey, Sjedinjene Američke Države. Iznosimo osnovne razlike u načinu života, ekonomiji, socijalnoj strukturi i osjećaju identiteta između povijesne (1951) i sadašnje (2001) populacije otoka. Primijenili smo suvremene metode (analize mikrosatelitnih DNA polimorfizama) kako bismo istražili utemeljenost lokalnih vjerovanja o izrazito visokom stupnju srođivanja i genetske homogenosti među stanovništvom otoka Suska. Analizom 18 markera na X-kromosomu u uzorku 36 osoba

procijenili smo kako prosječni koeficijent urođenosti populacije iznosi 5,7%, što je vrijednost blizu očekivane u potomaka prvih bratića. Analiza protezanja »linkage disequilibrium-a« (LD) u području Xq13–21 pokazala je kako su svi mogući parovi 10 mikrosatelitnih markera u ukupnom razmaku od 3.3 Mb bili u značajnom LD-u. Migracijska studija bila je dizajnirana kako bismo istražili učinke velikih promjena u okolišnim čimbenicima (Susak – SAD) i srođivanja (Susak – opća populacija Hrvatske) na neka biološki značajna kvantitativna svojstva, kao što su krvni tlak i masnoće u serumu. Rezultati su potvrdili povezanost između srođivanja i povišenog krvnog tlaka, koja je već i ranije bila opisana u svjetskoj literaturi. Posljednji ostaci tradicionalne otočne zajednice ubrzo će nestati.