Association among Education Level, Occupation Status, and Consanguinity in Tunisia and Croatia

Emna Kerkeni[1,2], Kamel Monastiri[3,4], Besma Saket[3,4], Diana Rudan[5], Lina Zgaga[6], Hassen Ben Cheikh[1,2]

[1]Genetics Laboratory, Medical Faculty of Monastir, Monastir, Tunisia
[2]Research Unit 02/UR/08-03, Medical Faculty of Monastir, Monastir, Tunisia
[3]Department of Pediatrics and Neonatology, EPS F. Bourguiba of Monastir, Monastir, Tunisia
[4]Research Unit 04/UR/08-19, Medical Faculty of Monastir, Monastir, Tunisia
[5]Holy Ghost General Hospital, Zagreb, Croatia
[6]Department of Medical Statistics, Epidemiology, and Medical Informatics, Andrija Štampar School of Public Health, Zagreb University School of Medicine, Croatia

> **Correspondence to:**

Hassen Ben Cheikh
Laboratoire de Génétique
Faculté de Médecine
Avenue Ibnou Sina
5019 Monastir. Tunisia
Hassen.BenCheikh@fmm.rnu.tn

> **Received:** July 12, 2006
> **Accepted:** July 20, 2006

> **Croat Med J. 2006;47:656-61**

Aim To investigate the association between education level, occupation status (a proxy for socio-economic status), and consanguinity in 2 large data sets from Tunisia and Croatia countries with different attitudes toward consanguinity.

Methods The sample of 1016 students, attending 5 university institutions in Monastir, Tunisia, were interviewed about the educational level and occupation status of their parents and the degree of parental relatedness. In Croatia, a sample of 1001 examinees from 9 isolated island populations was interviewed about their own educational level, occupation status, and consanguinity.

Results Prevalence of consanguinity (offspring of second cousins or closer) among 1016 Tunisian students was 20.1%, and 9.3% among 1001 Croatian isolates. In Tunisia, the association between consanguinity and both parental degree of education and parental occupation status was highly significant in women ($P<0.001$), but not significant in men. In Croatia, no statistically significant associations were noted, although there was a consistent trend of increased prevalence of consanguinity with lower education level or occupation status in both genders, but more pronounced in women.

Conclusion Association between education level, socio-economic status, and consanguinity needs to be taken into account in inbreeding studies in human populations. The relationship may be specific for each studied population and highly dependent on the cultural context. It is generally more pronounced among women in most settings.

Consanguineous marriages are unions between two persons who share at least one recent common ancestor (1). In clinical genetics, a consanguineous marriage is commonly defined as union between subjects related as second cousin or closer, equivalent to an inbreeding coefficient in their progeny of F≥0.0156 (2). This kind of union was known to increase the risk of homozygous recurrence of deleterious recessive genes (3,4), and this could explain the increase of polygenic or multifactorially determined diseases in populations with high prevalence of consanguinity (1,5,6). A number of studies reported that offsprings of consanguineous parents had higher rates of neonatal, post-neonatal, child, and infant mortality than those of non-consanguineous parents (7-11).

It is widely perceived that consanguinity is more prevalent among the underprivileged in the society (12-14). However, it is possible that factors that are not genetically determined, such as education level and socio-economic status of the subjects, have a confounding effect in the studies on consanguinity. To explore this, we investigated the association between education level, occupation status (a proxy for socio-economic status), and consanguinity in 2 large data sets available from Tunisia and Croatia. The analysis in these two countries with different attitudes toward consanguinity and causes and prevalence of inbreeding could reveal whether the presumed associations between education, occupation, and inbreeding could be generalized, or whether they are more complex and context-specific.

Participants and Methods

Tunisian sample

In Tunisia, we used a convenience sample of 1016 students attending five university institutions in Monastir. The students were men and women, aged 18-23 years, who agreed to participate in the survey. The study was conducted for 17 months, from January 2003 to May 2004. The students were surveyed about the educational level, profession, and the degree of biological relatedness of their parents. Students were considered to originate from consanguineous unions if their parents were related at the level of second cousins or closer. Paternal education was classified into 4 groups: 1) incomplete primary school or no education, 2) completed primary school, 3) completed secondary school, or 4) completed high school or more. Father's occupation status was classified into three groups: 1) professional (doctor, pharmacist, teacher, professor), 2) clerical, and 3) others (services, agricultural work, non-qualified employees, fisheries, day-laborers, not classifiable). Mother's occupation status was classified into three groups: 1) professional (doctor, pharmacist, teacher, professor), 2) housewife (women without employment outside of home), and 3) others (services, agricultural work, non-qualified employees).

Croatian sample

The field study that recruited 1001 examinees in Croatian island isolates was performed during 2002 and 2003 by a team from the Andrija Štampar School of Public Health, Zagreb University School of Medicine, and the Institute for Anthropological Research in Zagreb, Croatia. The details of field methods were described in detail elsewhere (15). Random samples of 100 individuals from 9 island settlements were collected. Sampling was based on computerized randomization of the most complete and accessible population registries in each village, which included medical records (Mljet and Lastovo islands), voting lists (Vis island), and household numbers (Rab island). Additional 101 examinees were recruited from the immigrants from all 9 villages who agreed to participate in the study, to form a genetically diverse control population that shared the same environment. Gender and age distribution in each sample and personal genetic histories of examinees (categorized as inbred, autoch-

thonous, admixed, and outbred) were presented elsewhere (15). An examinee in this sample was considered consanguineous when the same (non-marital) surname highly specific of the settlement, was found in at least one of parent of the student's father and mother. In such cases, further genealogical information was retrieved from parish registries in the 9 villages and the complete information on 3 ancestral generations for each individual included in the study allowed the assessment of the level of consanguinity. As in Tunisian sample, examinees were considered to originate from consanguineous unions if their parents were related at the level of second cousins or closer. The level of education of the examinees was assessed as the number of completed years in the school system, and their occupation was recorded in each case. The examinees were then categorized by both education level and occupation status, to match the categorization performed in Tunisian sample and ensure the comparability of results.

Statistical analyses

We first divided the Tunisian and Croatian sample into sub-samples defined by gender and by level of education or occupation status categories. Then, within each country and for each gender, we tested the null-hypothesis that the proportions of consanguineous individuals (found within these sub-samples) did not statistically differ between the sub-samples. This hypothesis was tested using χ^2 tests for independent samples and with appropriate number of degrees of freedom (3 for education level and 2 for occupation status).

Results

We compared the two samples at three levels (Tables 1 and 2): 1) education level, occupation status, and consanguinity between Tunisia and Croatia; 2) education level, occupation status, and consanguinity between men and women; and 3) association between consanguinity and education level/occupation status. The first analysis showed that consanguinity was two times more prevalent among Tunisian students (20.1%) than among Croatian remote island isolates (9.3%), whereas the level of education and

Table 1. Association between education level and consanguinity in the Tunisian and Croatian sample

	No. of consanguineous examinees/total number of examinees in each gender and education level category (%)*	
Education level	Tunisia	Croatia
Total:		
men	204/1016 (20.1)	43/455 (9.5)
women	204/1016 (20.1)	50/546 (9.2)
Less than primary school:		
men	15/73 (20.5)	6/38 (15.8)
women	48/181 (26.5)	13/82 (15.9)
Completed primary school:		
men	30/123 (24.4)	10/78 (12.8)
women	56/228 (24.6)	17/172 (9.9)
Completed secondary school:		
men	93/427 (21.8)	21/261 (8.0)
women	76/420 (18.1)	17/231 (7.4)
Completed high school:		
men	66/393 (16.8)	6/78 (7.8)
women	24/187 (12.8)	3/61 (4.9)
Statistics:		
men	$P=0.183$ ($\chi^2_1=4.85$)	$P=0.296$ ($\chi^2_1=3.70$)
women	$P<0.001$ ($\chi^2_1=36.32$)	$P=0.081$ ($\chi^2_1=6.74$)

*In Tunisia, the total number of examinees in each gender and education level category refers to parents of the examined students (thus the total number was n = 2×1016). In Croatia it refers to the examinees themselves (n = 1001).

Table 2. Association between occupation status and consanguinity in the Tunisian and Croatian sample

	Number of consanguineous examinees/ total number of examinees in each gender and occupation status category (%)*	
Occupation status	Tunisia	Croatia
Total:		
men	204/1016 (20.1)	43/455 (9.5)
women	204/1016 (20.1)	50/546 (9.2)
Professional:		
men	49/291 (16.8)	3/29 (10.3)
women	30/206 (14.6)	1/43 (2.3)
Clerical:		
men	36/181 (19.9)	1/28 (3.6)
women	–	3/74 (4.1)
Housewives:		
men	–	–
women	163/685 (23.8)	20/193 (10.4)
Agriculture, fishery, services, non-qualified employees, day laborers, and non-classifiable:		
men	119/544 (21.9)	39/398 (9.8)
women	11/125 (8.8)	26/236 (11.0)
Statistics:		
men	$P=0.223$ ($\chi^2_1=3.00$)	$P=0.546$ ($\chi^2_1=1.21$)
women	$P<0.001$ ($\chi^2_1=19.71$)	$P=0.109$ ($\chi^2_1=6.05$)

*In Tunisia, total number of examinees in each gender and occupation category refers to parents of the examined students (thus the total number was n = 2 × 1016), while in Croatia it refers to the examinees themselves (n = 1001).

occupational status was generally higher in Tunisia than in Croatia (Tables 1 and 2). The latter was expected given the nature of the samples of university students' parents in Tunisia vs remote islanders in Croatia. The second analysis showed that in both societies men were better educated than women. This was more pronounced in Tunisia, although among the professionals there were about as many (Tunisia) or more (in Croatia) women than men (Tables 1 and 2). In Tunisia, nearly 70% of examinees' mothers were housewives, compared with less than 40% housewives in the sample from Croatia.

Eight separate statistical tests were performed to analyze the association between education level and consanguinity in each gender (Table 1) and between occupation status and consanguinity (Table 2). In Tunisia, men with the highest education level and professionals had the lowest prevalence of consanguinity, but these differences were not statistically significant. Similar trends were observed for men in Croatia for their education level, with the decline in consanguinity prevalence from 15.8% to 7.8%, but not for occupation status. However, these differences did not reach statistical significance. For women in Tunisia, a strong trend of increasing prevalence of consanguineous marriages with decreasing education level and occupation status of student's mothers was noted (both $P<0.001$). In Croatia, similar trend was noted and was more apparent than in men (Tables 1 and 2), but the differences between groups did not reach formal statistical significance ($P=0.076$ for education level and $P=0.115$ for occupation status, respectively).

In Tunisia, 20.1% of consanguineous marriages in the sample was equivalent to a mean inbreeding coefficient α of 8.40×10^{-3}. In Croatia, this coefficient was two times lower. Among the related parents in Tunisia, 112 (54.9%) were related as first cousins. In Tunisia, 23.5% of women in consanguineous marriages had no education (ie, were illiterate), in comparison with 16.4% in non-consanguineous unions. In Croatia, both first-cousin marriages and illiteracy were extremely rare (under 3%). In Tunisia, the proportion of housewives was greater in the consanguineous group (79.9%) than that in the non-consanguineous group (64.3%), whereas the proportion of skilled women was higher in non-consanguineous group (21.7%) than in the consanguineous group (14.7%). Similar trends, although not as pronounced, were also observed in the Croatian sample.

Discussion

This study investigated the association between consanguinity, education level, and occupation status in two very different societies: the group of students in Tunisia and the isolates living on remote Croatian islands. There are different causes for the high prevalence of consanguinity in the two societies. In Tunisia, consanguinity is prevalent because of cultural factors, while in Croatia it occurs because of very limited mate choice on isolated and remote island communities. Still, the prevalence was twice as high in Tunisia as in Croatian islands, and in general population it would probably be even higher than in the sample of students. However, in Croatia the prevalence of consanguinity would be much lower in general population than among isolated islanders. It was, therefore, of interest to investigate whether the same associations between consanguinity, education level, and occupation status apply in both communities with prevalent consanguinity, but in entirely different contexts.

As the practice of consanguineous marriages is influenced by cultural, social, economic, religious, geographic, and demographic factors (16-18), some authors suggested that the highest rates of consanguineous unions were strongly associated with lower parental educational levels, marriage at an early age, low socio-economic status, illiteracy, and rural residence (19-24). This would, therefore, represent a major confounding effect in inbreeding studies conducted by genet-

ic epidemiologists. However, our study showed that this association, although present to some extent, is not of the scale that would affect results conducted in populations where consanguinity is prevalent solely because of isolation and limited mate choice. Even in countries where consanguinity is prevalent because of cultural practices, the association with education level and occupation status is mainly seen among women, but not in men. These findings have clear implications for design and conduct of genetic epidemiological studies that investigate the effects of consanguinity on human health. The observed associations seem to be considerably more pronounced in women, regardless of a society and cultural context. Associations between consanguinity and women's education level were reported in several other societies (20,24-27), although Hussain and Bittles could not confirm this negative correlation for women with primary school education (28). For men, the nature of association strongly depends on the cultural context, with reported examples ranging from negative correlation between inbreeding and social status (20,29), to a notion that more educated men were more likely to be married to cousins (27).

This study has several potential limitations. First, the sample in Tunisia is not likely to be representative of the general population, as university students were recruited and they are more likely to have parents of higher education and possibly also of higher socio-economical status. Second, consanguinity status, occupation status, and education level was not recorded for students themselves but for their parents. Therefore, an indirect measure of the relationship between consanguinity and education/occupation status was obtained, as the Tunisian sample only allowed an assessment of the likelihood of persons engaged in a consanguineous marriage (and who are not necessarily consanguineous themselves) to be better educated or of better occupation status. In Croatia, this measure is direct (all 3 variables are measured in each individual), but the sample is also not representative of the general population, but only of the isolate resource where limited mate choice, rather than culture or tradition, was a cause of inbreeding. Also, the data on consanguinity in the Croatian sample were verified in parish records, whereas the data on Tunisian sample were collected as the self-report survey. These problems limited cross-country comparisons, and they were therefore not performed. The differences across education and occupation status were investigated within countries and specific gender categories, which was still valid in both cases. Also, non-representativeness of both samples for general population may act in favor of the study design, as the analysis in these two samples from the populations with quite different attitudes toward consanguinity, causes and prevalence of inbreeding would allow us to assess whether the presumed associations between education, occupation, and inbreedi ng can be generalized, or are they more complex and context-specific.

The mean coefficient of inbreeding found in Tunisia ($8.4x10^{-3}$) was higher than that found in European and American populations, such as Netherlands ($0.1x10^{-3}$), Northeast of Uruguay ($1.7x10^{-3}$), or Northern Sweden ($2.0x10^{-3}$) (24-26). However, as hypothesized earlier, it was lower than in the majority of Arab and Muslim populations, eg, Egypt ($10.0x10^{-3}$), Turkey ($15.4x10^{-3}$), Oman ($17.6x10^{-3}$), and west India Muslims ($20.1x10^{-3}$) (30-33).

In conclusion, association between education level, socio-economic status, and consanguinity needs to be taken into account in inbreeding studies in human populations, and the relationship will often be highly specific for each studied population and strongly dependent on the cultural context.

Acknowledgments

This work was supported by the "Ministére Tunisien de l'Enseignement supérieur" by funds allocated to the research units 02/UR/O8-03 and 04/UR/08-19 and by the grant No. 0108330 from the Croatian Ministry of Science, Education, and Sports.

References

1 Rudan I, Rudan D, Campbell H, Carothers A, Wright A, Smolej-Narancic N, et al. Inbreeding and risk of late onset complex disease. J Med Genet. 2003;40:925-32. Medline:14684692

2 Modell B, Darr A. Science and society: genetic counselling and customary consanguineous marriage. Nat Rev Genet. 2002;3:225-9. Medline:11972160

3 Becker SM, Al Halees Z, Molina C, Paterson RM. Consanguinity and congenital heart disease in Saudi Arabia. Am J Med Genet. 2001;99:8-13. Medline: 11170087

4 Rittler M, Liascovich R, Lopez-Camelo J, Castilla EE. Parental consanguinity in specific types of congenital anomalies. Am J Med Genet. 2001;102:36-43. Medline: 11471170

5 Teebi AS, Teebi SA, Porter CJ, Cuticchia AJ. Arab genetic disease database (AGDDB): a population-specific clinical and mutation database. Hum Mutat. 2002;19:615-21. Medline:12007218

6 Rudan I, Smolej-Narancic N, Campbell H, Carothers A, Wright A, Janicijevic B, et al. Inbreeding and the genetic complexity of human hypertension. Genetics. 2003;163:1011-21. Medline:12663539

7 Sawardekar KP. Genetic analysis of lethal congenital malformations causing perinatal mortality at Nizwa Hospital, Oman. Clin Genet. 2004;66:239-43. Medline: 15324324

8 Bittles AH, Grant JC, Shami SA. Consanguinity as a determinant of reproductive behaviour and mortality in Pakistan. Int J Epidemiol. 1993;22:463-7. Medline: 8359962

9 Grant JC, Bittles AH. The comparative role of consanguinity in infant and childhood mortality in Pakistan. Ann Hum Genet. 1997;61:143-9. Medline: 9177121

10 Stoltenberg C, Magnus P, Skrondal A, Lie RT. Consanguinity and recurrence risk of stillbirth and infant death. Am J Public Health. 1999;89:517-23. Medline:10191794

11 Jorde LB. Consanguinity and prereproductive mortality in the Utah Mormon population. Hum Hered. 2001;52:61-5. Medline:11474206

12 Fuster V, Colantonio SE. Socioeconomic, demographic, and geographic variables affecting the diverse degrees of consanguineous marriages in Spain. Hum Biol. 2004;76:1-14. Medline:15222677

13 Hussain R, Bittles AH. Sociodemographic correlates of consanguineous marriage in the Muslim population of India. J Biosoc Sci. 2000;32:433-42. Medline:11075637

14 Liascovich R, Rittler M, Castilla EE. Consanguinity in South America: demographic aspects. Hum Hered. 2001;51:27-34. Medline:11096268

15 Rudan I, Biloglav Z, Vorko-Jovic A, Kujundzic-Tiljak M, Stevanovic R, Ropac D, et al. Effects of inbreeding, endogamy, genetic admixture, and outbreeding on human health: a "1001 Dalmatians" study. Croat Med J. 2006;47:601-10. Medline: 16909458

16 Khlat M. Consanguineous marriages in Beirut: time trends, spatial distribution. Soc Biol. 1988;35:324-30. Medline: 3241995

17 Hussain R. Community perceptions of reasons for preference for consanguineous marriages in Pakistan. J Biosoc Sci. 1999;31:449-61. Medline:10581876

18 Fuster V, Colantonio SE. Consanguinity in Spain: socio-economic, demographic, and geographic influences. Hum Biol. 2002;74:301-15. Medline:12030656

19 Khlat M. Consanguineous marriage and reproduction in Beirut, Lebanon. Am J Hum Genet. 1988;43:188-96. Medline:3400644

20 Khoury SA, Massad D. Consanguineous marriage in Jordan. Am J Med Genet. 1992;43:769-75. Medline:1642259

21 al Husain M, al Bunyan M. Consanguineous marriages in a Saudi population and the effect of inbreeding on prenatal and postnatal mortality. Ann Trop Paediatr. 1997;17:155-60. Medline:9230979

22 Hussain R, Bittles AH. Consanguineous marriage and differentials in age at marriage, contraceptive use and fertility in Pakistan. J Biosoc Sci. 1999;31:121-38. Medline:10081242

23 Alper OM, Erengin H, Manguoglu AE, Bilgen T, Cetin Z, Dedeoglu N, et al. Consanguineous marriages in the province of Antalya, Turkey. Ann Genet. 2004;47:129-38. Medline:15183745

24 McCullough JM, Rourke DH. Geographic distribution of consanguinity in Europe. Ann Hum Biol. 1986;13:359-67. Medline:3767309

25 Lusiardo A, Barreto I, Hidalgo PC, Bonilla C, Bertoni B, Portas M, et al. Consanguinity in two Uruguayan cities: historical evolution and characteristics (1800-1994). Ann Hum Biol. 2004;31:513-25. Medline:15739381

26 Bittles AH, Egerbladh I. The influence of past endogamy and consanguinity on genetic disorders in northern Sweden. Ann Hum Genet. 2005;69:549-58. Medline:16138913

27 Jurdi R, Saxena PC. The prevalence and correlates of consanguineous marriages in Yemen: similarities and contrasts with other Arab countries. J Biosoc Sci. 2003;35:1-13. Medline:12537152

28 Hussain R, Bittles AH. The prevalence and demographic characteristics of consanguineous marriages in Pakistan. J Biosoc Sci. 1998;30:261-75. Medline:9746828

29 Gunaid AA, Hummad NA, Tamim KA. Consanguineous marriage in the capital city Sana'a, Yemen. J Biosoc Sci. 2004;36:111-21. Medline:14989534

30 Hafez M, El-Tahan H, Awadalla M, El-Khayat H, Abdel-Gafar A, Ghoneim M. Consanguineous matings in the Egyptian population. J Med Genet. 1983;20:58-60. Medline:6842535

31 Donbak L. Consanguinity in Kahramanmaras city, Turkey, and its medical impact. Saudi Med J. 2004;25:1991-4. Medline:15711682

32 Rajab A, Patton MA. A study of consanguinity in the Sultanate of Oman. Ann Hum Biol. 2000;27:321-6. Medline:10834296

33 Bittles AH, Hussain R. An analysis of consanguineous marriage in the Muslim population of India at regional and state levels. Ann Hum Biol. 2000;27:163-71. Medline: 10768421