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Orofacial analysis on the Adriatic islands: an epidemiological study of malocclusions on Hvar Island

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SUMMARY The prevalence of malocclusion was studied on the Hvar island, Croatia. This island is characterized by high rates of endogamy, inbreeding and reproductive isolation. The sample for this study comprised 224 children, 126 males and 98 females from all schools on the island of Hvar (20.2 per cent of the total public school population). The sample was selected according to age, sex distribution, and demographic characteristics. The Angle Classes, overjet, overbite, crowding, spacing, and the type of anterior bite were examined.

Class I buccal relationship was found in 47.3 per cent of the subjects, Class II in 45.1 per cent, and Class III in 5.4 per cent. Normal overjet was the most frequent finding (69.9 per cent). Normal overbite was seen in 42.5 per cent, deep in 49.1 per cent, and very deep in 2.7 per cent of the cases. Open bite was diagnosed in seven subjects (3.1 per cent) and edge-to-edge bite in six (2.7 per cent). The frequency of crowding was 57.1 per cent, normal relationships were observed in 34.9 per cent, and spacing in 8 per cent.

The results of this investigation showed that the inhabitants of Hvar, who are characterized by a high rate of inbreeding and traditional diet, have more Class II malocclusions, deep bites, and midline shifts than the general Croatian population but not higher than other modern urbanized groups. Since the lack of chewing stress in this population cannot be considered as the main explanation for the above phenomenon, the genetic influence on the development of these occlusal traits in the inbred population of Hvar requires further investigation.

Introduction

Analysis of the prevalence of occlusal traits in isolated human populations can provide valuable information regarding the aetiology of malocclusions and other complex traits (Rudan *et al*., 1999). The term 'malocclusion', encompassing all deviations of the teeth and jaws from normal alignment, includes a number of distinct conditions, including discrepancies between tooth and jaw size (crowding and spacing); malrelationships of the dental arches (sagittal, transverse, and vertical) and malpositioning of individual teeth (Lombardi and Bailit, 1972).

In traditional human societies, which are characterized by genetic uniformity, malocclusions are quite rare (Corruccini and Pacciani, 1989), while increased incidence of malocclusion can be found in the modern industrialized world (Kelley and Harvey, 1977; Proffit, 1999). Research has suggested that secular increases in malocclusions have accelerated during the last 150 years in technologically advanced communities, after having shown relatively modest changes for 6000 years (Andrik, 1963; Corruccini and Lee, 1984; Vyslozil and Jonke 1994; Weiland *et al*., 1997). This is mostly explained by a shift to a less tough diet associated with the exertion of less force on the jaws during chewing (Corruccini and Lee, 1984).

On the other hand, some authors suggest that a high prevalence of malocclusions may be found in isolated populations (Maatouk *et al*., 1995; Ben-Bassat *et al*., 1997) and that effects of inbreeding can be observed in the orofacial system (Goldhamer *et al*., 1990; Zlotoroga, 1997). The main genetic consequence of inbreeding is an increase in the proportion of homozygotes and recessive genes (Bodmer and Cavalli-Sforca, 1976). If some recessive genes are mutated and/or responsible for some disturbances in occlusal relationships, prevalence of such traits is expected to be higher in an inbred group than in the general population. In highly outbred groups such as the US population, a greater rate of malocclusions can be observed (Proffit, 1999), perhaps due to widening of the gene pool and/or dietary changes.

The purpose of this study was: (1) to evaluate the prevalence of some occlusal traits and malocclusions among the population of Hvar Island, Croatia; (2) to determine whether some of the occlusal traits are more prevalent compared with the general Croatian population; (3) to evaluate the age group that is critical in the development of occlusion; and (4) to provide the

basis for future studies of genetic influence on the prevalence and progression of malocclusions.

Subjects and methods

The study sample encompassed 224 children from all schools on the island of Hvar, Croatia, representing 20.2 per cent of the island's population for the same age. The sample was targeted in accordance with age, sex distribution, and demographic characteristics of all school children on the island of Hvar (Tables 1 and 2).

All children were of school age (7–14 years) with a mean age of 131.5 months (SD 28.1). Eighteen children had orthodontic treatment and their data were registered from their medical records.

Upper and lower alginate impressions were taken and poured in dental stone. The same investigator made all the recordings. The values of overjet, overbite, and midline shifts were measured using a calliper (Digimatic, Mitutoyo Corporation, Japan) with digital output, accurate to the nearest 0.01 mm.

To score buccal sagittal relationship, Angle's classification was used. According to the registration method developed by the World Health Organisation and the Federation Dentaire International (Siriwat and Jarabak, 1985), the mesial and distal antero-posterior molar relations were divided in two categories: cusp-to-cusp < one cusp width, and one cusp width and more.

Table 1 Age distribution of the sample.

Age (years)	Total children	Sample	Sample proportion of total (%)
7	129 (11.6%)	27 (12%)	20.9
8	153 (13.8%)	34 (15.2%)	22.2
9	125 (11.3%)	28 (12.5%)	22.4
10	117 (10.6%)	21 (9.4%)	17.9
11	143 (12.9%)	26 (11.6%)	18.2
12	136 (12.3%)	27 (12.1%)	19.9
13	168 (15.1%)	28 (12.5%)	16.7
14	138 (12.4%)	33 (14.7%)	23.9
Total	1109 (100%)	224 (100%)	20.2

The variables used in this study were divided into four main groups according to Ben-Bassat *et al*. (1997).

Antero-posterior dimension

Molar relationship: Angle's Class I, II, or III. Categories of the antero-posterior molar relationship were subselected to: normal, distal, cusp-to-cusp < one cusp width; distal, one cusp width and over; mesial, cusp-to-cusp < one cusp width; mesial, one cusp width and more; and unrecordable.

Overjet. This was evaluated to the nearest 0.5 mm. Normal range was determined at 0.5–4.0 mm.

Anterior crossbite. This included mandibular overbite and edge-to-edge position.

In Angle's classification, half unit Class II or half unit Class III were considered Class II or Class III, respectively (Ben-Bassat *et al*., 1997).

Transversal dimension

Discrepancy between the upper and lower arch midline. Posterior crossbite: buccal/palatal; teeth in an edge-to-edge position were included.

Vertical dimension

Overbite, including edge-to-edge and open bites. Normal range was determined at 0.5–3.5 mm, deep at 3.6–6.5 mm, and very deep > 6.5 mm.

Other

Crowding as well as *spacing* were also registered according to (1) no crowding/no spacing (including crowding ≤ 0.5 mm and spacing 0.5 mm), (2) crowding (> 0.5 mm), and (3) spacing (> 0.5 mm). Both maxilla and mandible were calculated together. Median diastema (≥ 1 mm).

The criteria for defining normal occlusion were a Class I canine and molar relationship, a positive overjet up to 4 mm, and overbite up to 3.5 mm, and well aligned

Table 2 Sex and demographic distribution.

	Inhabitants under 15 years of age			Sample		
	Males	Females	Total	Males	Females	Total
Towns	766	711	1477 (69.6%)	86	68	154 (68.8%)
Villages	339	305	644 (30.4%)	40	30	70 (31.2%)
Total	1105 (52.1%)	1016 (47.9%)	2121	126 (56.3%)	98 (43.7%)	224

There are three towns on the island of Hvar: Hvar, Starigrad, and Jelsa. These towns are administrative centres for a number of villages around the island and the majority of inhabitants live in them.

arches (Brin *et al*., 1996). Slightly irregular arches and crowding or spacing up to 0.5 mm in the upper or lower arch were included within normal limits.

Twelve casts were measured twice by the author. Measurement error was ranked from 0.01 mm to a maximum of 0.04 mm. For Angle Classes, crossbites, crowding, and spacing no error was found in the repeat series of evaluation.

Statistical analyses were performed using 'Statistica 5.1 '97 Edition'. Sexual dimorphism was evaluated by the chi-square test (Angle Classes; anterior crossbite; normal, deep, and very deep bite; midline shift, crowding and spacing, medium diastema) and the *t*-test was performed for independent samples (overjet, overbite).

Results

The total prevalence of malocclusion among the sample was 93.3 per cent; in boys 94.4 per cent and 91.8 per cent in girls according to the criteria of Brin *et al*. (1996) for defining normal occlusion. The only sex-related significant difference was found for overjet (mean value 3.68 ± 1.88 mm for males and 3.22 ± 1.51 mm for females, $P < 0.05$). The occlusal parameters are presented in Table 3 and the antero-posterior molar relationship in Table 4.

Antero-posterior dimension

In the total sample, Class I was found in 106 children (47.3 per cent), Class II in 101 children (45.1 per cent), and Class III in 12 children (5.4 per cent). Five children (2.2 per cent) had unrecordable molar relationships. Table 3 shows the distribution of the molar relationship categories on both sides. Symmetrical Angle Classes were observed in 151 children. The most frequent symmetrical Angle Class was Class I (43.8 per cent), followed by Class II (22.8 per cent) and Class III (0.9 per cent).

Table 3 Distribution of occlusal traits.

Occlusal traits		*n*	%
Antero-posterior dimension	Anterior crossbite	2	0.9
Transversal dimension	Midline shift	101	45.1
	Right bucal crossbite	20	8.9
	Right lingual crossbite	1	0.4
	Left bucal crossbite	24	10.7
	Left lingual crossbite	1	0.4
Vertical dimension	Open bite	7	3.1
	Edge-to-edge anterior bite	3	1.3
Other	Crowding	128	57.1
	Spacing	18	8.0
	Midline diastema	29	12.9

A normal overjet (0.5–4.0 mm) predominated among the children examined with a frequency of 69.6 per cent and anterior crossbite was observed in 1 per cent of cases. The mean value of the total sample was 3.48 ± 1.74 mm.

Transversal dimension

The distributions of midline shifts and buccal and lingual crossbites are presented in Table 4. The mean value of midline shifts was 1.58 mm (SD 0.71 mm). A small midline shift (0.1–1 mm) was found in 25 children (12.1 per cent), moderate (1.1–2 mm) in 57 (27.7 per cent), and large (>2 mm) in 19 (9.2 per cent). It can be seen that among the total midline shifts ($n = 101$) more than half were moderate (56.4 per cent), small shifts were noted in 24.8 per cent and large shifts in 18.8 per cent of cases.

A buccal crossbite was more frequent than a lingual crossbite and was observed in 8.9 per cent of subjects on the right side and in 10.7 per cent on the left. Bilateral crossbite was noted in 10 subjects (4.5 per cent).

Vertical dimension

A normal overbite (<3.5 mm) was seen in 42.5 per cent, deep (3.6–6.5 mm) in 49.1 per cent, and very deep (>6.5 mm) in 2.7 per cent of the cases. Seven subjects (3.1 per cent) had an open bite and six (2.7 per cent) an edge-to-edge vertical bite. The mean overbite value of the total sample was 3.49 ± 1.67 mm; for females 3.35 ± 1.61 mm, and for males 3.60 ± 1.71 mm.

Other

The frequency of crowding was 57.1 per cent, normal relationships were found in 34.9 per cent, and spacing in 8 per cent of the sample. A midline diastema was observed in 12.9 per cent.

Table 4 Antero-posterior molar relationship.

Angle	Category	Right molar		Left molar	
		n	%	*n*	%
Class I	Normal	125	55.8	134	59.8
Class II	Distal: cusp-to-cusp < one cusp width	52	23.2	42	18.8
	Distal: ≥ one cusp width	28	12.5	31	13.8
Class III	Mesial. cusp-to-cusp < one cusp width	5	2.2	2	0.9
	Mesial: ≥ one cusp width	3	1.3	4	1.8
Unrecordable		11	4.9	11	4.9
Total		224	100	224	100

Discussion

The population of Hvar represents one of the last genetic isolates in Europe. Since 1972, continuing interdisciplinary investigation of the rural population of the island has included the study of biomedical, sociocultural, biocultural, and genetic traits. (Rudan, 1972, 1975; Rudan *et al*., 1982a, b, 1986; Janićijević, 1985; Smolej, 1987; Šimić and Rudan, 1990; Šimić *et al*., 1992; Roguljić *et al*., 1997; Sujoldžić, 1997; Martinović *et al*., 1998; Waddle *et al*., 1998). Many of these analyses have clearly demonstrated the division of the island villages into two clusters—western and eastern. However, analysis of the prevalence of caries has not shown any difference between the eastern and western parts of the island (Lauc *et al*., 2000). The importance of environmental factors in the aetiology of caries is a probable explanation. In this respect, the prevalence of malocclusion on the island of Hvar is of special interest because more hereditary factors contribute to the incidence of malocclusion than to caries. Of note are some population structure studies (Rudan *et al*., 1990; Roguljić *et al*., 1997) of Hvar that have indicated exceptionally high rates of inbreeding, kinship, endogamy, and isonymous marriages (marriages between individuals sharing the same surname). The frequency of isonymous marriages in some villages is as high as 0.39 and the coefficient of kinship (the probability of two individuals having genes in common by the same descent) is as high as 0.10. The coefficient of kinship for the whole island is 0.041; the only higher coefficient ever reported is 0.092 in Amish, Nebraska, USA (Rudan, 1997). A coefficient of endogamy (both parents born in the same settlement) of 0.755 and the proportion of isonymous marriages (5–35 per cent; Roguljić *et al*., 1997) contributed to the findings of the high rate of reproductive isolation.

The strategy designed by Siriwat and Jarabak (1985) and Ben-Bassat *et al*. (1997) was followed with minimal modifications. The most important results in the sample of Hvar's children were that crowding predominated (57.1 per cent), deep bite was found in 49.1 per cent of the cases, and normal overjet in 69.6 per cent. A high rate of midline diastema was observed (45.1 per cent). The total prevalence of malocclusions was 93.3 per cent, and among the children a similar frequency of Class I and Class II malocclusions was found. The results show that the rate of occurrence of Class II malocclusions was 45.1 per cent—which is almost equal to the findings in the genetically isolated populations of Ashkenazi Jews (46.4 and 46.2 per cent, respectively; Ben-Bassat *et al*., 1997). Although the incidence of Class II malocclusions in the present investigation is in agreement with those authors, it was higher than recorded in the data from different Croatian areas (Table 5). Compared with the data from the American, Asian, Australian, and European populations (Lombardi and Bailit, 1972), the Hvar sample also showed a relatively high prevalence of Class II malocclusions. It is important to stress the finding that in more than one-third of subjects with a Class II malocclusion, this was one cusp width or more (Table 4) because it represents a greater pathological pattern. This could be explained by a genetic influence on the incidence of Class II malocclusions due to endogamy and inbreeding. Other investigators have found that the incidence of Class I buccal relationship varies from 63.2 per cent in the coastal area, middle Dalmatia (Radica-Sorić, 1978), to 75.8 per cent in the northern part, Slavonia (Ceranić, 1977), Class II from 19.3 per cent in Slavonia (Ceranić, 1977) to 29.9 per cent in middle Dalmatia (Radica-Sorić, 1978), and Class III from 3.3 per cent in the other coastal area, the peninsula of Istria (Cipruš, 1982), to 5.9 per cent in middle Dalmatia (Radica-Sorić, 1978). Open bite varies from 0.8 to 5 per cent and crowding from 55 per cent on the northern Adriatic coast to 65 per cent on the Istria peninsula (Cipruš, 1982).

The 5.4 per cent prevalence of a Class III malocclusion seen in this study coincides with the range from 1.8 to 5.9 per cent in other parts of the country (Table 5), as well as with the findings from other populations (Lombardi and Bailit, 1972; Proffit, 1999). Obviously, inhabitants of the island do not have greater hereditary predisposition to this anomaly and genetic uniformity did not increase the frequency of Class III malocclusions.

In this study, a relatively high frequency of deep bite (49.1 per cent) could be associated with the high incidence of Class II malocclusions.

It is obvious that several genetic and environmental interacting factors are related to the aetiology of malocclusions. Soft diet, mouth breathing, tongue trusting, sleeping posture, sucking, and other habits as well as specific factors (skeletal growth disturbances, muscle dysfunction, disturbances in embryologic and dental development) interact with heredity in the development of major types of malocclusion (Profitt, 1999). The difficulty in separating these factors is

Table 5 Sagittal plane relationship of the dental arches in different Croatian regions.

Author	Region	Class I (%)	Class II (%)	Class III (%)
Ceranić, 1977	Slavonia	75.8	19.3	5.4
Cipruš, 1982	Istria and northern Adriatic coast	73.5	20.9	3.3
Radica-Sorić, 1978	Middle Dalmatia	63.2	29.9	5.9
This study	Island of Hvar, Dalmatia	47.6	45.2	4.8

obvious: in terms of gene–environment interactions, intraoral environmental change may be a decisive factor, but this change may also reveal previously masked genetic effects (Corruccini, 1991). The National Health and Nutrition Survey III undertaken in the United States between 1989 and 1994 showed a frequency of crowding from 42.3 per cent (age 8–11) to 54.5 per cent (age 12–17), Class II malocclusions from 55.1 per cent (age 8–11) to 67.7 per cent (age 12–17), and Class III from 2.9 per cent (age 8–11) to 5.7 per cent (age 12–17; Proffit, 1999), which is not very different from the frequencies observed in this investigation. However, the US population is uniform in dietary habits, which are dependent on industrially processed foods (Corruccini, 1991), while the inhabitants of Hvar preserve a more traditional diet consisting of home-produced foods providing consistent loading during mastication.

On the basis of the results of Ben-Bassat *et al.* (1997), as well as of the current study, it may be stated that isolated population groups have behavioural and/or genetically dependent parameters different from those in the general populations, but not much greater than the crowding and Class II and III malocclusions revealed in the US data. It should also be noted that although the population of Hvar is isolated and characterized by a distinct lifestyle, high prevalence of malocclusion, typical for developed countries, is observed and the overall frequency of malocclusion of 93.3 per cent seen in this study is even higher than the data from some western countries (Egermark-Eriksson, 1982).

Conclusions

The results of this investigation show that the inhabitants of Hvar with a high rate of inbreeding and traditional diet have more Class II malocclusions, deep bites, and midline shifts than the general Croatian population but not more than other modern urbanized groups. Since the lack of chewing stress in this population cannot be considered as the main explanation for the above phenomenon, it is proposed that the genetic influence on development of these occlusal traits in the inbred population of Hvar should be the subject of future investigations. Such studies should investigate the prevalence of different occlusal traits in clusters of different inbreeding rates with the same environmental conditions in the same population. The findings of the current study may have possible clinical implications in the prevention of malocclusion in Hvar.

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