Stomatologija, Baltic Dental and Maxillofacial Journal, 8:53-6, 2006

Craniofacial morphology in parents of cleft children and healthy individuals

Erika Nagle, Uldis Teibe, Ieva Balode

SUMMARY

Craniofacial morphology with respect to orofacial clefts has been widely studied. Objective of this study was to determine distinct craniofacial parameters in parents who have cleft children. Materials and methods. Craniofacial anthropometric measurements (total) have been studied in 57 cleft fathers, 67 cleft mothers, 39 control males, and 38 control females. All parameters were compared between cleft parents and control (for males and females separately). Results. Statistical analysis showed significant differences ($p<0.05$) between the cleft parents and the controls for 18 measurements characterizing head, face, orbital, nasal, and oral region. Conclusions. Results of this study suggest that craniofacial morphology in parents of children with clefts is distinctive from that observed in healthy individuals in Latvia population. Such data could be used in evaluation persons at risk for having cleft child, and also to define an anthropometric evaluation system for parents who have cleft children.

Grant: Taiwan - Baltic joint research project "Identification of Genes Involved in Craniofacial Morphogenesis and Susceptibility to Orofacial Clefting in Human Genome Scan".

Key words: anthropometry, craniofacial parameters, orofacial clefts.

INTRODUCTION

This study was designed to determine deviations in craniofacial morphology among parents of cleft children. Distinct craniofacial features in phenotypically unaffected parents have been reported [1]. The parental craniofacial form represents the hereditary influences on the craniofacial form of their offspring [2]. Studying craniofacial morphology of parents who have children with orofacial clefts could provide effective tool in understanding predisposition to clefts and therefore could give significant input in preventive activities. Relevant anthropometric features can be used to identify individuals at greater risk to have "cleft" genes and consequently to be at greater risk for producing a child with a cleft. Anthropometrical approach was used to evaluate craniofacial morphology. Several studies [3, 4, 5] have shown an association between specific craniofacial parameters and the presence of orofacial clefts in their children. Anthropometric features distinguishing various races/ ethnic groups are reported [6]. For this reason it seems very important evaluate deviations in anthropometric parameters of the individuals from the same population. The aim of this study was to determine are there differences in craniofacial morphology between parents who have children with clefts and individuals without family history of orofacial clefting in Latvia population.

Erika Nagle *- Dr. Biol., assoc. prof., Department of Medical Biology and Genetics, Riga Stradins University, Latvia*
Uldis Teibe *- Dr. Biol., prof., Department of Physics, Riga Stradins University, Latvia*
Ieva Balode *- M.D., clinical geneticist, Clinic of Medical genetics, Children`s Clinical University Hospital*

Address correspondence to Erika Nagle, Department of Medical Biology and Genetics, Riga Stradins University, Latvia, Dzirciema iela 16, Riga, LV-1007 Latvia.
E-mail address: erika.nagle@rsu.lv

MATERIALS AND METHODS

In this cross – sectional study the subjects were parents of children with cleft lip/palate (cleft fathers, cleft mothers) born in Latvia, and clinically healthy Latvia residents (control). A total 201 subjects (57 cleft fathers, 67 cleft mothers, 39 control males, and 38 control females) were included in our study. Craniofacial measurements from control group were taken if an individual met such criteria: Latvia resident, normal craniofacial configuration, no known history of craniofacial abnormalities in the family. Criterion for cleft parents – child born with cleft lip alone, cleft lip and palate, cleft palate alone. The individuals of both groups (cleft parents and control individuals) were informed about the nature and aim of the study, and informed consent was obtained. All measurements were carried out with the GPM Anthropological Instruments, Siber Hegner & Co. AG. The measurements were taken with a sliding calliper, spreading calliper, and measuring tape. Measurements of cleft parents were performed at the Institute of Stomatology Riga Stradiòð University, measurements of control individuals at the Riga Stradins University and at the University of Daugavpils. Measurements (total 20) with easily defined landmarks that encompass all major areas of the head and face were included (Table 1). In identification of landmarks and for anthropometric measuring techniques we have followed instructions described by Kolar JC, Salter ME [7]. Because sexual dimorphism in Latvia residents was observed in almost all parameters that include craniofacial region [8], measurements of this study were evaluated separately for males and females in both cleft and control group. Descriptive and inferential statistics have

Table 1. Craniofacial landmarks and measurements

Landmark	Measurement	Landmark	Measurement
eu – *euryon*	eu-eu – maximum head breadth	ex – *exocanthion*	ex-ex – biocular width
ft – *frontotemporale*	ft-ft – minimal frontal breadth		Interpupillary distance
t – *tragion*	t-t – cranial base width	n – *nasion* sn – *subnasale*	n-sn – nose height
g – *glabella* op – *opistocranion*	g-op – maximum head length	al – *alare*	al-al – nose width
g – *glabella* op – *opistocranion*	g-op – head circumference		columella width
zy – *zyguon*	zy-zy – maximum facial breadth	sn – *subnasale* ls – *labiale superius*	sn-ls – philtrum length
go – *gonion*	go-go – mandible breadth	*cph – cristae philtri*	cph-cph – philtrum width
tr – *trihion* gn – *gnation*	tr-gn – physiognomial face height	ch – *cheilionn*	ch-ch – labial fissure width
n – *nasion* gn – *gnation*	n-gn – morphological face height	ls – *labiale superius* sto – *stomion*	ls-sto – upper vermilion height
en – *endocanthion*	en-en – intercanthal width	sto – *stomion* li – *labiale inferius*	sto-li – lower vermilion height

been used for interpretation of the results. All the measurements were processed by SPSS (11.0). Mean, standard deviation, and independent sample *t* test were used for evaluating the difference between cleft parents and controls. For data normal distribution study groups were studied by using Kolmogorov-Smirnov Z test.

RESULTS

Craniofacial anthropometric measurements of 201 subjects were obtained. All measurements are given in centimetres. Mean values, standard deviation, two-tailed significance, and mean differences of the measurements are shown in tables 2 and 3. Statistically significant differences between the cleft parents and the controls

($p< 0.05$) for 18 measurements characterizing head, face, orbital, nasal, and oral region were obtained. Table 2 shows anthropometric measurements of the craniofacial region of cleft fathers and control males. Higher values in cleft fathers when compared with control males were observed in several parameters commonly used to characterize the head and face. Cleft group had wider and longer heads (measurements *eu-eu; g-op*), greater head circumference (*g-op1*), wider faces *(zy-zy*), and wider mandibles

Table 2. Comparison of craniofacial measurements (cm) between cleft fathers and control males

	Clefts (n = 57)		Control (n = 39)			
	Mean	SD	Mean	SD	MD	p
eu-eu	15.97	0.52	15.42	1.01	0.55	0.01*
ft-ft	11.04	0.48	11.49	1.10	-0.46	0.01*
t-t	14.55	0.70	14.42	0.59	0.13	0.36
g-op	19.60	0.68	19.31	0.68	0.29	0.04*
g-op 1	58.16	1.43	57.37	1.49	0.79	0.01*
zy-zy	14.36	0.68	13.31	0.98	1.06	0.01*
go-go	11.19	0.83	10.54	0.63	0.65	0.01*
tr-gn	18.59	1.03	18.73	0.73	-0.14	0.46
n-gn	12.19	0.70	12.41	0.60	-0.23	0.11
en-en	2.83	0.30	2.91	0.30	-0.08	0.21
ex-ex	8.53	0.47	10.63	0.58	-2.10	0.01*
pupils	6.11	0.41	6.39	0.41	-0.28	0.01*
n-sn	5.23	0.56	5.87	0.54	-0.65	0.01*
al-al	3.54	0.36	3.53	0.32	0.01	0.92
columella	0.66	0.13	1.04	0.18	-0.38	0.01*
sn-ls	1.46	0.28	1.27	0.23	0.20	0.01*
cph-cph	1.42	0.32	1.08	0.21	0.33	0.01*
ch-ch	5.10	0.71	5.08	0.37	0.02	0.88
ls-sto	0.76	0.34	0.81	0.23	-0.05	0.43
sto-li	0.77	0.24	1.07	0.26	-0.31	0.01*

* mean differences are statistically significant

Table 3. Comparison of craniofacial measurements (cm) between cleft mothers and control females

	Clefts (n = 67)		Control (n = 38)			
	Mean	SD	Mean	SD	MD	p
eu-eu	15.04	0.62	14.58	0.59	0.46	0.01*
ft-ft	10.62	0.51	10.66	0.74	-0.04	0.74
t-t	13.60	0.70	13.55	0.50	0.05	0.68
g-op	18.41	0.68	18.33	0.66	0.08	0.55
g-op 1	55.52	1.60	55.22	1.87	0.31	0.38
zy-zy	13.37	0.65	12.24	0.80	1.13	0.01*
go-go	10.27	0.53	9.69	0.75	0.58	0.01*
tr-gn	17.29	0.82	17.70	0.79	-0.42	0.01*
n-gn	11.01	0.53	11.76	0.62	-0.75	0.01*
en-en	2.81	0.25	2.66	0.24	0.15	0.01*
ex-ex	8.20	0.37	10.06	0.60	-1.85	0.01*
pupils	5.85	0.30	6.04	0.42	-0.19	0.01*
n-sn	4.84	0.32	5.67	0.57	-0.83	0.01*
al-al	3.24	0.26	3.28	0.27	-0.03	0.55
columella	0.59	0.13	0.88	0.13	-0.29	0.01*
sn-ls	1.24	0.25	1.14	0.19	0.10	0.04*
cph-cph	1.16	0.21	0.91	0.18	0.25	0.01*
ch-ch	4.85	0.42	4.65	0.34	0.20	0.01*
ls-sto	0.70	0.18	0.78	0.14	-0.08	0.01*
sto-li	0.71	0.20	1.03	0.14	-0.33	0.01*

* mean differences are statistically significant

(*go-go*). Cleft fathers showed longer and wider philtrum (*sn-ls, cph-cph*). However some other measurements in cleft fathers showed lower values in comparison with controls. The minimal frontal breadth (*ft-ft*) of the cleft fathers was significantly lower than for the controls. Similarly, the biocular width (*ex-ex*), interpupillary distance, nose height (*n-sn*), columella width, lower vermilion height (*sto-li*) of the cleft fathers was lover than those of controls. Several other craniofacial measurements in cleft fathers and in controls did not show significant differences. Cranial base width (*t-t*), face height (*tr-gn, n-gn*), intercanthal width (*en-en*), nose width (*al-al*), and upper vermilion height ((*ls-sto*) showed no significant differences between cleft fathers and control males.

Table 3 shows anthropometric measurements of the craniofacial region of cleft mothers and control females. The results of these measurements in respect to head (*eu-eu*) and mandible breadth (*go-go*), philtrum length (*sn-ls*), and philtrum width (*cph-cph*) are similar to those obtained in males. They showed significantly higher values for the cleft group when compared to the controls. Cleft mothers showed also wider mouth (*ch-ch*) and greater intercanthal width (*en-en*) than those of controls. Like in male group other parameters in cleft group showed lower values when compared with controls. Cleft mothers had reduced facial height both physiognomial (*tr-gn*) and morphological (*n-gn*). Comparing measurements from the orbital, nose, and mouth region we found that biocular width (*ex-ex*), interpupillary distance, nose height (*n-sn*), columella width, upper (*ls-sto*) and lower(*sto-li*) vermilion height was smaller in cleft mothers. Although deviations from controls, except of head width, were not observed in head and face region. Minimal frontal breadth *(ft-ft*), cranial base width (*t-t*), head length (*g-op*), head circumference (*g-op1*), and facial breadth (*zy-zy*) were not significantly different between two groups. No differences were observed in nose width (*al-al*) as well. In general our results showed that almost all measurements differed significantly between the two groups – cleft parents and controls.

DISCUSSION

The anthropometric contribution of characteristics craniofacial morphology in parents of cleft children has been focus of research for a decade of years. Many researchers have evaluated craniofacial structures in cleft parents. Differences among their techniques of measurements and methodology of interpretation their results limit the possibility of comparison results. Despite this several authors obtained similar results, and overall results appear to support the hypothesis that parents of children with non-syndromic clefts tend to differ from the general population in certain craniofacial parameters. The measurements selected for the study were intended to develop the picture of craniofacial morphology in cleft parents and control individuals of the same population. Measurements were taken in an attempt to establish the main facial parameters of Latvia residents. Our results presented in tables 2 and 3 supports data observed by other authors [9, 10,] that craniofacial measurements are different in parents who have orofacial cleft children in comparison with normal control. In our study reduced parameters in cleft parents (both fathers and mothers) were observed in orbital, nasal, and oral region. Contrary parents of children with clefts compared to the control group have increased parameters in the head width as well as philtrum width and length. Longer philtrum was observed also in more severe affected patients when compared to less severe affected patients with cleft lip [10]. Thickness of upper lip also coincidence greatly between cleft parents (in our study thinner upper lip was observed in cleft mothers) and cleft patients [12]. This could suggest that long philtrum and thin upper lip could be considered as microform of the clefts. Our results with respect to head width are contradictory with those observed by some other researches. As it was reported [10] head breadth in cleft parents is reduced when compared with controls, but our results showed statistically significant increase of this parameter in both cleft fathers and cleft mothers. Results of anthropometric measurements in respect to mandible breadth in our study showed similar results as in other authors report [10,13]. The mandible breadth of the cleft parents was significantly higher than for the controls. However, some other authors [9] have reported lower values of this measurement in cleft mothers in comparison with control females. Some researches [10] have reported smaller facial dimensions in mothers of cleft children, and it was reported also that individuals at risk for having child with orofacial clefts can be recognized through reduced facial height [3]. Results of our study concerning facial dimensions are in agreement with this report. We have observed reduced facial height in mothers, who have children with clefts.

CONCLUSIONS

- Results of this study support hypothesis that craniofacial morphology in parents of children with orofacial clefts is different from that observed in healthy individuals in Latvia population.
- Data about anthropometric features in parents of cleft children could be useful to define an anthropometric evaluation system for parents who have cleft children.

ACKNOWLEDMENTS

We wish to thank all participants, especially parents of cleft patients, as well as students of the University of Daugavpils and Riga Stradins University, for participation, patience and support.

REFERENCES

1. Perkiomaki MR, Yoon YJ, Tallents RH, Barillas I, Herrera-Guido R, Moss ME. Association of Distinct Craniofacial features in Nonsyndromic Cleft Lip and Palate Family members. *Cleft Palate Craniofac J* 2003; 40: 397-402.
2. McIntyre GT, Mossey PA. The craniofacial morphology of the parents of children with orofacial clefting: a systematic

review of cephalometric studies. *J Orthod* 2002; 29 :23-9.
3. Ward RE, Bixler D, Jamison PL. Cephalometric evidence for a dominantly inherited predisposition to cleft lip-cleft palate in a single large kindred. *Am J Med Genet* 1994; 50: 57-62.
4. Mossey PA, Arngrimsson R, McColl J, Vintiner GM, Connor JM. Prediction of liability to orofacial clefting using genetic and craniofacial data from parents. *J Med Genet* 1998; 35: 371-8.
5. Yoon YJ, Perkiomaki MR, Tallents RH, Barillas I, Herrera-Guido R, Fong CT, et al. Transverse craniofacial features and their genetic predisposition in families with nonsyndromic unilateral cleft lip and palate. *Cleft Palate Craniofac J* 2004; 41: 256-61.
6. Farkas LG, Katic MJ, Forrest CR, Alt KW, Bajic I, Baltadjiev G, et al. International anthropometric study of faciak morphology in various ethnic groups/races. *J Craniofac Surg* 2005; 16: 615-46.
7. Kolar JC, Salter ME. Craniofacial anthropometry: practical measurement of the head and face for clinical, surgical and research use. Sprinfield, IL: Charles C Thomas publisher; 1997.
8. Nagle E, Teibe U, Ka¸oka Dz. Craniofacial anthropometry in a group of healthy Latvian residents. *Acta Med Lithuanica* 2005; 12: 47-53.
9. Suzuki A, Takenoshita Y, Honda Y, Matsuura C. Dentocraniofacial Morphology in Parents of Children with Cleft Lip and/or Palate. *Cleft Palate Craniofac J* 1999; 36: 131-8.
10. ALEmran SE, Fatani E, Hassanain JE. Craniofacial variability in parentsofchildren with cleft lip and cleft palate. *J Clin Pediatr Dent* 1999; 23: 337-41.
11. Yeow VK, Huang MH, Lee ST, Fook Chong SM. An anthropometric analysis of indices of severity in the unilateral cleft lip. *J Craniofac Surg* 2002; 13: 68-74.
12. Vegter F, Hage JJ. Facial Anthropometry in Cleft Patients: A Historical Appraisal. *Cleft Palate Craniofac J* 2001; 38: 577-81.
13. Mossey PA, McColl J, O‘ Hara M. Cephalometric features in the parents of children with orofacial clefting. *Br J Oral Maxillofac Surg* 1998; 36: 202-12.

Received: 01 03 2006
Accepted for publishing: 27 06 2006