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ORIGINAL ARTICLE

Treatment Outcome Based on GOSLON Score of Multiple Factors in Unilateral Cleft Lip and Palate Children of Four Different Ethnic

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Abstract

Objective: To evaluate and compare the effect of each congenital and postnatal treatment factors in treatment outcome based on dental arch relationship (DAR) of four different populations at a time using GOSLON yardstick. **Material and Methods:** 432 unilateral cleft lip and palate subjects (Japanese, Malay ethnic group, Bangladeshi and Pakistani populations) age 5 to 12 years were taken before orthodontic treatment and alveolar bone grafting. The DAR was assessed by GOSLON Yardstick. Independent t-test was performed to compare the GOSLON score for each factors. A multiple comparison (Anova) was also conducted between the GOSLON score of four different populations. The significance level was set at $p<0.05$. **Results:** Statistically no significant different was found among the congenital factors. However, the family history of Class III malocclusion showed most likely to associate with, though there was no significant difference ($p=0.069$). Significant difference revealed between two techniques of cheiloplasty in both Malay ethnic group and Pakistani population ($p=0.038$ and $p=0.000$, respectively). Gender and Techniques of palatoplasty also showed significant difference in Pakistani population ($p=0.026$ and $p=0.000$, respectively). Japanese and Bangladeshi population showed no significant differences. Also no significant differences found between the GOSLON score and different countries. **Conclusion:** The treatment outcome based on DAR significantly varies in individual population [Malay ethnic group (cheiloplasty); Pakistan (gender, cheiloplasty and palatoplasty)].

Keywords: Congenital Abnormalities; Jaw Abnormalities; Cleft Palate; Cleft Lip.

Introduction

Cleft deformities remain a significant and interesting challenge for the medical fraternity. WHO recommendations promoting the cleft lip and palate (CLP) as the burden of global initiation because of an increase in life expectancy. The prevalence and incidence of CLP varies in different population, race and ethnic background [1]. Beside the aesthetic problem, psychological dilemma and variety of functional problems like feeding, speech, hearing problem can emerge to a cleft patient. Dental irregularities are also observed frequently among these patient compared to the others [2].

Treatment of CLP patient is quite complex and lengthy procedure with undergoing surgical and nonsurgical multidisciplinary treatments from birth to adulthood. When a patient born with CLP, a number of surgery take place in 1st two years [3,4]. Maxillary growth retardation is often observed in patients with repaired unilateral cleft lip and palate (UCLP) which finally leads to Class III malocclusion, concave facial profile and mid facial growth deficiency. According to previous literature, various factors including both congenital and post natal treatment factors are responsible for this maxillary growth retardation which later on go towards the unfavorable treatment outcome of a Unilateral Cleft Lip and Palate (UCLP) patient [5-8].

In recent years, multitude of research has been done worldwide on treatment outcome of CLP patient using different index [2,4,5,9-18] Among them GOSLON yardstick is the most commonly used index. The widespread use of GOSLON yardstick has proved itself as reliable, robust, sound index in evaluation treatment outcome based on dental arch relationship [19]. To the best of our knowledge, all of those researches have been ended with total GOSLON score. The GOSLON score of individual factor has not yet been evaluated. Moreover, most of the studies were on particular center or on single based population.

For the first time, herein, we have retrospectively evaluated the effect of each congenital and postnatal treatment factors on treatment outcome of four different populations at a time using GOSLON yardstick.

Material and Methods

Subjects

Total 432 UCLP subjects of different populations: Japanese, Malay ethnic group, Bangladeshi and Pakistani populations were included in this study before any orthodontic treatment and bone grafting. Age of the subjects was ranged from 5 to 12 years and all of them underwent different techniques of cheiloplasty and palatoplasty. However, subjects with any associated anomalies or syndromes were excluded from the study. Demographic information of all the participants from four different populations have presented on Table 1.

One hundred and forty Japanese UCLP subjects were included from the records of orthodontic clinic of Hokkaido University Hospital. The mean age of the subjects was 6.85 ± 1.56. The Malaysian sample was matched for inclusion and exclusion criteria with a sample of 107

subjects, collected from the records of Hospital Universiti Sains Malaysia and the mean age was 7.69 ± 2.46. Eighty-four Bangladeshi and 101 Pakistani UCLP subjects were collected from the archives of two individual renowned hospitals from two different countries. The mean age was 7.69 ± 2.46 and 8.05 ± 0.79 for Bangladeshi and Pakistani UCLP subjects respectively. Appropriate sample size calculation of each country had been calculated separately using PS software.

Table 1. Distribution of subjects according to demographic and clinical characteristics.

Variables		Population			
		Japanese	Malay Ethnic Group	Bangladeshi	Pakistani
Gender	Male	76	63	43	58
	Female	64	44	41	43
UCLP Side	Right	38	37	33	35
	Left	102	70	51	66
UCLP Types	Complete	97	91	31	101
	Incomplete	43	16	53	0
F/H of Cleft	Positive	20	20	50	30
	Negative	120	87	34	71
F/H of C-III	Positive	24	N/A	34	N/A
	Negative	116		50	
Cheiloplasty	MMVF	80	66	35	65
	MMT	60	41	49	36
Palatoplasty	PBT	127	38	44	70
	P2	13	69	40	31
Upper LI on Cleft Side	Present	75	15	N/A	N/A
	Missing	65	92		
Wearing Psot	Nothing	72	N/A	N/A	N/A
	Wear Plate	68			

F/H: Family History; C-III: Class III Malocclusion; MMVF: Modified Millard with Vomer Flap; MMT: Modified Millard Technique; MT: Millard Technique; PBT: Push Back Technique; P2: Two Stage Palatoplasty; LI: Lateral Incisor; Psot: Pre Orthopedic Surgical Treatment; N/A: Not applicable.

Different treatment protocols (wearing post, timing and techniques of surgery) have been observed for the management of UCLP among four different countries. The treatment protocol of UCLP children of each country have shown in Table 2.

Table 2. Treatment protocol of different populations.

Population		Treatment				
		Cheiloplasty	Palatoplasty	Wearing Psot	Alveolar Bone Grafting	Any Orthodontic Treatment
Japan	Age (Average) Technique	5 Months 1. Modified Millard 2. MMVF	20 Months 1. Push Back 2. P2	✓	X	X
Malaysia	Age (Average) Technique	5 Months 1. Millard 2. Modified Millard	18 Months 1. Bardach 2. Von Langenbeck	X	X	X
Bangladesh	Age (Average) Technique	5 Months 1. Millard Technique 2. Modified Millard	18 Months 1. Bardach 2. V-Y push Back	X	X	X
Pakistan	Age Technique	6-10 Months 1. Millard 2. Modified Millard	12-18 Months 1. Von Langenbeck 2. V-Y Push Back	X	X	X

MMVF: Modified Millard with Vomer Flap; P2: Two Stage Palatoplasty.

Intra- and Inter-Examiner Agreements

Three examiners rated all the Japanese UCLP subjects and five examiners rated all the Malay ethnic group, Bangladeshi and Pakistani UCLP subjects with two weeks interval. The kappa scores for the GOSLON Yardstick showed good to very good intra- and inter-examiner agreements for all the populations. The kappa score ranged from 0.654 to 0.909 and 0.738 to 0.898 for the intra- and inter-examiner agreements in Japanese population [5]; 0.890 to 0.933 and 0.768 to 0.960 for the intra- and inter-examiner agreements in Malay ethnic group [16]; 0.856 to 0.904 and 0.809 to 0.951 for the intra- and inter-examiner agreements in Bangladeshi population [12] and 0.773 to 0.849 and 0.685 to 0.861 for the intra- and inter-examiner agreements in Pakistani population [9].

Assessment

GOSLON Yardstick was used to evaluate dental arch relationship based on each individual factor. According to GOSLON Yardstick, five categories are rated: 1: excellent; 2: good; 3: fair; 4: poor; 5: very poor which reflect a growth range of dental arch relationship. Group 1 (excellent), a favorable relationship, shows advantageous skeletal form, with a positive overjet and overbite. Patients exhibit an Angle Class II division 1 malocclusion in this group. Straightforward orthodontic treatment or no treatment need at all in this group. Group 2 (good) is also a favorable relationship with Class I dental relationship and also indicates straightforward orthodontic treatment. Group 3 (fair) presents as an edge-to-edge dental relationship where patient need of more complex orthodontic treatment to correct the Class III malocclusion and other possible arch deformities, but a good result can still be predictable. Group 4 (poor), an unfavorable facial growth with reverse overjet of 3-5 mm, which indicates the limits of orthodontic treatment, may require an orthognathic procedure. Group 5 (very poor) represents a significant skeletal Class III relationship with mandatory surgical correction [20].

Statistical Analysis

Independent t-test was performed among to compare the GOSLON score for each congenital and post natal treatment factors for all populations. Anova test was also conducted to observe the multiple comparison between the GOSLON score of four different populations. These analyses were carried out using the statistical package SPSS Version 24.0 (SPSS Inc., Chicago, IL, USA). The significance level was set at $p<0.05$.

Ethical Aspects

This study was conducted under taking the proper ethical clearance by the institutions (Jouf University, Universiti Sains Malaysia, Fukuoka Dental College and Hokkaido University).

Results

Japanese Population

Table 3 shows mean, standard deviation, 95% CI and p-value of the each factors congenital factors including male-female, side of cleft, type of cleft, family history of cleft, family history of Class III and presence of upper lateral incisor of cleft side. Statistically no significant different was found among the congenital factors. However, the family history of Class III malocclusion showed most likely to associate with, though there was no significant difference (p=0.069).

No significant difference concerning the postnatal treatment factors in Japanese UCLP subjects was observed (Table 3). Millard technique of cheiloplasty (GOSLON score = 2.9833) presented better prognosis compare to modified Millard technique with vomer flap (GOSLON score = 3.2000).

Malay Ethnic Group

Though no significant differences had been observed among any congenital factors but the GOSLON mean score of all the factors showed more than 3 which indicates fair to poor treatment outcome among the subjects. The subjects who operated by Millard technique of cheiloplasty, was noticeably different with the subjects who operated by modified Millard technique (p=0.038). However, palatoplasty showed no significant differences (Table 3).

Bangladeshi Population

No significant differences were found in score for individual congenital factors. However, the subjects who had family history of Class III malocclusion showed greater score (GOSLON score = 3.4706) than the subjects who did not have any family history of Class III malocclusion (GOSLON score = 3.0800) (p=0.068) of family history of Class III malocclusion also indicated that there were some differences though it was not significant (Table 3). No significant difference was found in score for postnatal factors among Bangladeshi UCLP subjects.

Pakistani Population

Independent t test demonstrated significant differences in GOSLON score for male and female. The p value was 0.026. Male (GOSLON score = 2.8103) presented better prognosis compare to Pakistani female UCLP subjects (GOSLON score = 3.3721) (Table 3).

The result shows significant differences with both palatoplasty (p=0.000) and cheiloplasty (p=0.000). The mean GOSLON score of the V-Y pushback palatoplasty was 4.0323, which indicates very poor treatment outcome compared to the subjects who treated with Von Langenbeck technique of palatoplasty (mean GOSLON score = 2.6143). These findings imply heightened risk for unfavorable DAR among Pakistani UCLP subjects. Likewise, according to mean GOSLON score, results also identified that the subjects operated by Millard technique were more likely than the subjects operated by modified Millard technique (Table 3).

Table 3. GOSLON score of Japanese, Malay ethnic group, Bangladeshi and Pakistani populations.

Variables	Category	N	Mean	SD	95%CI		p-value
					Lower	Upper	
GOSLON Score of Japanese Population							
Gender	Male	76	3.0526	0.79824	-0.38544	0.14696	0.377
	Female	64	3.1719	0.78790			
Side	Right	38	3.0789	0.53935	-0.33768	0.26028	0.798
	Left	102	3.1176	0.87080			
Type	Complete	97	3.1237	0.80697	-0.23419	0.34208	0.712
	Incomplete	43	3.0698	0.76828			
F/H of Cleft	Positive	20	2.9500	0.75915	-0.56212	0.19545	0.340
	Negative	120	3.1333	0.79846			
F/H of C-III	Positive	24	3.3750	0.71094	-0.02536	0.67191	0.069
	Negative	116	3.0517	0.80049			
Upper LI on Cleft Side	Present	75	3.0659	0.85378	-0.39585	0.16037	0.404
	Missing	65	3.1837	0.66688			
Wearing Psot	Nothing	72	3.1111	0.77923	-0.27425	0.25791	0.952
	Wear Plate	68	3.1029	0.81295			
Palatoplasty	PBT	127	3.1181	0.80291	-0.33967	0.57589	0.611
	P2	13	3.0000	0.70711			
Cheiloplasty	MMVF	80	3.2000	0.86273	-0.48291	0.04958	0.110
	MM	60	2.9833	0.67627			
GOSLON Score of Malay Ethnic Group							
Gender	Male	63	3.0469	0.76490	-0.42287	0.19104	0.456
	Female	44	3.1628	0.81446			
Side	Right	37	3.1892	0.73929	-0.46232	0.16965	0.361
	Left	70	3.0429	0.80642			
Type	Complete	91	3.0989	0.80353	-0.38667	0.45947	0.865
	Incomplete	16	3.0625	0.68007			
F/H of Cleft	Positive	20	3.1000	1.07115	-0.37901	0.39510	0.967
	Negative	87	3.0920	0.70928			
Upper LI on Cleft Side	Present	15	3.1333	0.74322	-0.48091	0.38816	0.833
	Missing	92	3.0870	0.79355			
Palatoplasty	BT	38	3.0263	0.67731	-0.41879	0.21055	0.513
	VLT	69	3.1304	0.83864			
Cheiloplasty	MT	66	2.9697	0.76399	-0.62701	-0.01897	0.038*
	MMT	41	3.2927	0.78243			
GOSLON Score of Bangladeshi Population							
Gender	Male	43	3.3488	0.92282	-0.19159	0.64536	0.284
	Female	41	3.1220	1.00487			
Side	Right	33	3.2424	0.93643	-0.43846	0.42420	0.974
	Left	51	3.2353	0.99173			
Type	Complete	31	3.3548	1.14159	-0.24963	0.61968	0.400
	Incomplete	53	3.1698	0.84889			
F/H of Cleft	Positive	50	3.3200	1.01900	-0.22451	0.62921	0.348
	Negative	34	3.1176	0.87956			
F/H of C-III	Positive	34	3.4706	0.96091	-0.02992	0.81109	0.068
	Negative	50	3.0800	0.94415			
Palatoplasty	BT	44	3.2273	0.80301	-0.44449	0.39903	0.915
	V-Y PB	40	3.2500	1.12660			
Cheiloplasty	MMT	49	3.2619	0.91223	-0.46615	0.45662	0.984
	MT	35	3.2571	1.12047			

GOSLON Score of Pakistani Population							
Gender	Male	58	2.8103	1.24896	0.06867	1.05483	0.026*
	Female	43	3.3721	1.21544			
Side	Right	35	3.0571	1.25892	-0.51370	0.53708	0.965
	Left	66	3.0455	1.27015			
F/H of Cleft	Positive	30	2.9667	1.12903	-0.42880	0.66448	0.670
	Negative	71	3.0845	1.31743			
Palatoplasty	V-Y PB	31	4.0323	0.79515	0.95549	1.88045	0.000*
	VLT	70	2.6143	1.18313			
Cheiloplasty	MT	65	2.7077	1.16891	0.47325	1.44470	0.000*
	MMT	36	3.6667	1.19523			

*Statistically Significant.

In Table 4, the result of multi population comparison between the Malaysian, Japanese, Bangladeshi and Pakistani UCLP subjects are presented. Though no significant differences was observed between the countries, but the mean GOSLON score of each individual factors of Malaysian, Japanese and Bangladeshi UCLP subjects was more than 3 which specifies that the treatment outcome was towards the unfavorable.

Table 4. Multi population comparison of GOSLON score.

Comparison	MD	SE	95%CI		p-value
			Lower	Upper	
Japan Vs. Malaysia	0.01368	0.12233	-0.3106	0.3379	1.000
Japan Vs. Bangladesh	-0.13095	0.13148	-0.4795	0.2176	1.000
Japan Vs. Pakistan	0.05764	0.12437	-0.2720	0.3873	1.000
Malaysia Vs. Bangladesh	-0.14464	0.13888	-0.5127	0.2235	1.000
Malaysia Vs. Pakistan	0.04395	0.13217	-0.3064	0.3943	1.000
Bangladesh Vs. Pakistan	0.18859	0.14068	-0.1843	0.5615	1.000

MD: Mean Difference; SE: Standard Deviation.

Discussion

According to literature, CLP has been accepted as one of the most common congenital anomalies in the head and neck region worldwide. The global prevalence of CLP is 1:600 live births [14]. Moreover, the incidence among Asian population is reported as approximately 1.30:1000 live births [1]. Regarding non-syndromic clefts 1.41:1000 in Japanese [5], 1:941 in Malaysian [11], 3.9:1000 in Bangladeshi [12], 1:523 Pakistani population have been documented [9].

Over the last two decades, GOSLON Yardstick index is observed as the most commonly used index. In this prime study, we assessed total 432 UCLP subjects of different populations: Japanese (n=140), Malay ethnic group (n=107), Bangladeshi (n=84) and Pakistani (n=101) populations for evaluation of dental arch relationship based on each congenital and post natal treatment factors using GOSLON Yardstick. GOSLON Yardstick proved itself as reliable, valid able, robust, golden index for categorizing dental arch relationships and interfering of facial morphology outcomes. Not only that, it also can predict surgical outcomes at early age of patients as well as can predict outcome based on the individual factors as well as results of cephalometric analysis [7].

In earlier study, the treatment outcome based on dental arch relationship of Japanese (5), Malay ethnic group [16], Bangladeshi [12] and Pakistani [9] UCLP children following cheiloplasty and palatoplasty and also explored the various congenital (UCLP type, UCLP side, family history of cleft, family history of Class III, presence of upper lateral incisor on cleft side) and post natal treatment [pre orthopedic surgical treatment (Psot), cheiloplasty, palatoplasty] factors that affects dental arch relationship of UCLP children. According to previous studies, the total mean GOSLON scores of Japanese, Malay ethnic group, Bangladeshi and Pakistani populations were 3.11, 3.15, 3.238, and 3.04 respectively, representing the fair to poor treatment outcome [5,9,12,16].

Moreover, palatoplasty was significantly correlated with dental arch relationship in Japanese population [5]. In Bangladeshi population, family history of Class III malocclusion showed significant correlation with dental arch relationship [12]. In addition, gender and both cheiloplasty, palatoplasty were significantly correlated with dental arch relationship in Pakistani population [9]. However, Malay ethnic group did not show any significant associations [16]. In recent times, numerous researches took place in treatment outcome of UCLP children using GOSLON Yardstick in different population all over the world. Surprisingly, all the researches ended up with total mean GOSLON score. Figure 1 will give an idea about the total mean GOSLON score in different populations globally.

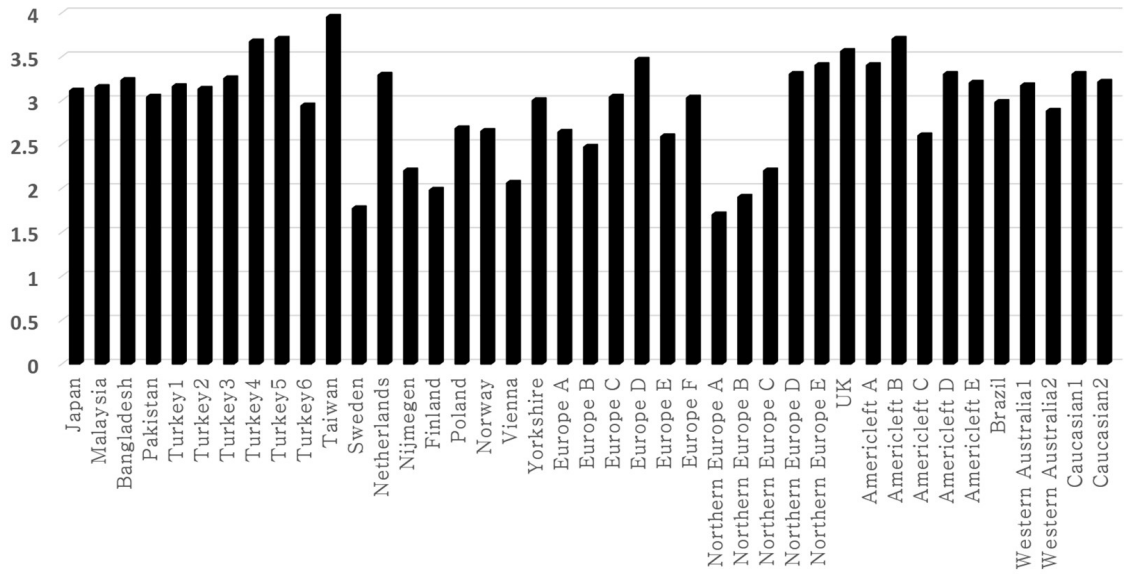

Figure 1. Global mean GOSLON score in different populations.

Novelty of this research, (1) total 432 subjects are included in this study which is noticeably higher than the number of the subjects of previous studies, (2) we know of no studies with individual GOSLON score of all of the congenital and post natal treatment factors that affect the treatment outcome of UCLP children till to date. This is necessary to know that which factor is affecting the treatment outcome thus clinicians can take proper steps or precaution or modify their treatment plan at the early age of the patient. For the very first time, this study evaluated the effects of each

individual factors, (3) as a final point, this study also presented a retrospective evaluation of treatment outcome among four different populations together as well as the comparison of the treatment outcome of four different populations of UCLP children for the first time in all over the world.

In this study, our result showed, techniques of cheiloplasty had significant differences in Malay ethnic group and Pakistani population. Both population underwent Millard technique and modified Millard technique of cheiloplasty. Millard technique (Malay ethnic group: p=0.038; Pakistani population: p=0.000) had significantly better maxillary growth in compared to modified Millard technique of cheiloplasty. This difference may be due to unfavorable growth pattern in the modified Millard technique, which accredit to the tension developed, which finally leads to rotation development. The greater lip tension is predicted to cause mainly dentoalveolar constriction rather than skeletal changes [21]. Nevertheless, in previous studies, skeletal changes involving anterior portion of maxilla in antero-posterior and transverse dimension also reported [22,23]. Similar studies were also carried out using 5 year olds' index [24]. However, lip length was not considered in present study, which could justify the use of modified Millard technique.

In Pakistani population, techniques of palatoplasty also showed significant differences in the treatment outcome of UCLP children. The disadvantageous effects of palatoplasty on growth have been widely reported previously [5,11,14,25]. Consistent comparative results of different methods of palatoplasty are occasionally reported. A large variation in the sample type and numerous confounding factors, such as size of defect, extent of defect, timing of repair and most importantly growth response makes assessment very difficult. Findings of our models of Pakistani population suggested that the VY pushback technique had significantly unfavorable treatment outcome. This is may be due to excessive scar tissues formation and the undermining of soft tissue during palatal repair, which finally lead to unfavorable maxillary growth [26].

However, in some previous studies, use of Von-Langenbeck technique also reported better outcomes due to lower scar formation [26,27]. Interestingly, in earlier study, V-Y pushback technique was also significantly correlated with unfavorable treatment outcome compared to Bardach technique in Bangladeshi population [12]. Similarly findings are also reported in Japanese population from another earlier study [5]. However, in our present study, palatoplasty did not remain a significant precise factor among Japanese, Bangladeshi and Malay ethnic group.

This study observed a frequent incidence of occurring CLP in male children among all the populations which is in covenant and also the evidence of findings of previous epidemiological study of cleft worldwide [1]. Surprisingly, female UCLP children of Pakistani population revealed significantly unfavorable treatment outcome. Similar findings also described in a Chinese population [1]. However, the etiology behind this finding is so far blurred.

The impetus for the present study came from the observation of comparing the treatment outcome among four different populations for the first time. However, no significant differences found in treatment outcome based on individual factors among different populations. This findings

may be due to all of their treatment protocol is some extend similar or may be all of their total outcomes were towards the fair to poor as we reported in earlier studies.

Conclusions

1. No significant difference was found in Japanese population in UCLP children using GOSLON Yardstick;
2. Significant differences were found between techniques of cheiloplasty and unfavorable dental arch relationship among UCLP children of Malay ethnic group using GOSLON Yardstick;
3. No significant difference was found in Bangladeshi population in UCLP children using GOSLON Yardstick;
4. Significant differences were found among the gender, techniques of cheiloplasty, and technique of palatoplsty with unfavorable dental arch relationship in UCLP children of Pakistani population using GOSLON Yardstick;
5. No significant difference was found in the treatment outcomes among Japanese, Malay ethnic group, Bangladeshi and Pakistani populations using GOSLON Yardstick.

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Conflict of Interest: The authors declare no conflicts of interest.

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