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Research Article

A CROSS SECTIONAL DESCRIPTIVE STUDY ON COMPARATIVE EVALUATION OF DENTAL ARCH DIMENSIONS IN THALASSEMIC AND NORMAL CHILDREN

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ABSTRACT

Introduction: Beta thalassemia or "Thalassemia Major" is a common genetic blood disorder with known skeletal and craniofacial deformities that may result in cosmetic abnormalities and emotional distress in these developing children. Present investigation attempted with the aim to compare dental arch dimensions in thalassemic and normal children (age group 7-14 years) of central India population including the effect of systemic disease on general and orofacial growth in thalassemic patients versus normally growing children.

Methods: A random sample of 100 children was equally divided in two groups as thalassemic and normal to investigate the longitudinal, horizontal and vertical dental arch dimensions in thalassemic children and compare them with corresponding values in a group of unaffected normal children. Alginate impressions were made to make standardized dental casts. Various maxillary and mandibular arch dimensions such as interincisal width, intercanine width, interpremolar width and intermolar width were measured on maxillary and mandibular dental casts using electronic digital sliding vernier calliper with conventional linear measurement method. Error estimation was done using Dahlberg's formula and Student's t-test was used to compare the results.

Results: The results of the study indicate that, thalassemic children had statistically significant decreased arch width compared to healthy children ($p < 0.05$). They also had decreased maxillary arch length (inter-incisor 27.34 ± 2.41, inter-canine 35.52 ± 2.82, inter-premolar 41.49 ± 4.02 and inter-molar 50.92 ± 2.73 respectively) whereas mandibular arch length was unaffected.

Conclusion: Thalassemic children had narrow maxilla and mandible with thin cortical plates as compared to systemically healthy children.

INTRODUCTION

Thalassaemia is a group of inherited defects in the synthesis of either the α or β polypeptide chains of haemoglobin, referred to as α and β thalassaemia, respectively. Based on genetic and clinical entities, thalassaemia are classified as homozygous, heterozygous, or compound heterozygous. The homozygous form of β-thalassaemia (thalassaemia major) exhibits the most severe clinical symptoms with marked orofacial defects. β-thalassaemia is transfusion-dependent and is commonly manifested during the first year of life. Frequently, affected infants are severely anaemic, fail to thrive and gain weight normally, and become progressively paler. Diarrhoea, recurrent fever, susceptibility to infection, hepatosplenomegaly, and retardation of growth are common presenting symptoms.[1]

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Thalassemia Major (TM) is a serious systemic disorder produces skeletal and craniofacial deformities affecting the aesthetics of affected patients as compared to their normal healthy peers.[2] Patients show spacing and forward drift of maxillary incisors for which orthodontic treatment may be indicated.

There are few reports in literature about the dimensional characteristics of the dental arches that may affect aesthetics, oral hygiene, caries risk, gingival and periodontal health and overall quality of life of the affected patients.[3-5] Many studies have investigated arch dimensional changes in various stages of the growth and development period in normally growing children.[6-8] Certain studies are done in thalassemic which are focussed on evaluation of soft tissue profiles and uvuloglossopharyngeal dimensions using cephalometric analyses. Amini *et al*[9] compared soft-tissue profile characteristics of patients with beta thalassaemia major (BTM) and non-thalassaemic subjects with a similar skeletal pattern in age group of 9-11 years using cephalograms and concluded that horizontal soft-tissue measurements were larger relative to non-thalassemic subjects that might have implications for their future orthodontic or orthognathic management. In the study of evaluation and comparison of uvuloglossopharyngeal dimensions (UGDs) in thalassemic and normal children [10], certain UGDs like tongue length, vertical distance between hyoid bone and C3-RGN line and distance between hyoid bone and mandibular plane were significantly shorter in thalassemic patients ($P < 0.01$), except for the middle air way space which was wider ($P < 0.01$) in thalassemic children. These findings also might have implications in orthodontic treatment of such patients.

Regarding the arch dimensions on concrete dental casts, no studies are carried out in thalassemic and other systemically affected patients. Therefore, the present investigation attempted to study the important factors related to arch width, arch length and arch depth including the effect of systemic disease on general and orofacial growth in thalassemic patients versus normally growing children and the precautions to be taken during orthodontic treatment.

MATERIAL AND METHODS

Sample selection

Following the institutional ethical committee approval, oral examination was carried out with written informed consent obtained from all the parents**.** Sample size estimate was obtained based on the study by Al-Wahadni *et al*. (2005).[11] Based on the data presented, for an effect size of 0.57, a sample of 100 subjects (50 in each group) provided this magnitude of difference of measurement with 95% CI and 80% power. Accordingly, the randomised sample consisted of 100 subjects in the age range of 7-14 years and divided into two groups. Study group consisted of 50 children previously diagnosed with Thalassemia major (TM) reporting for regular blood transfusion in the collaborated medical institutes. Control group consisted of 50 healthy children without any systemic or oral disease selected randomly from outpatient department. Children showing clinically evident interproximal dental caries, oligodontia, tooth anomalies, oral habits, facial deformity and those undergoing or underwent orthodontic treatment were excluded from the study. The mean age of control sample was 10.92 ± 2.39 years and that of diseased group was 11.06 ± 2.41 years. Fifty children of study group were previously diagnosed with Thalassemia major (TM) and used to report for regular blood transfusion in the collaborated medical institutes all of which were government organizations situated at Nagpur and Wardha where patients were drained from all areas in the territories of the central India. All these patients were from low socioeconomic status and in need of regular blood transfusions who reported to the collaborated government institutes where blood transfusion was provided to them free of cost.

A particular age group of 7-14 years was selected which consisted of both mixed and permanent dentition for measuring and comparing the dental arch form in different parameters. The mixed dentition stage of development corresponds to the time when early orthodontic therapy might be started in patients with severe malocclusion while permanent dentition corresponds to the time when full conventional orthodontic therapy can be started and thus the preferred age group was considered to be the favourable age for orthodontic intervention.

Preparation of dental models

Alginate impressions of maxillary and mandibular arches were made replicating all minor details of oral cavity and total 200 standardized dental models /casts were made with dental stone.

Arch dimensions

All measurements of arch dimensions namely arch width; arch length and arch depth were measured by conventional linear measurement method[12] using electronic digital sliding vernier calliper twice on the casts at two weeks interval by the same operator. The arch width was measured in three segments as interincisal width, intercanine width, interpremolar width and intermolar width. (Fig 1a and 1b) Arch length was measured by combining anterior and posterior arch length.

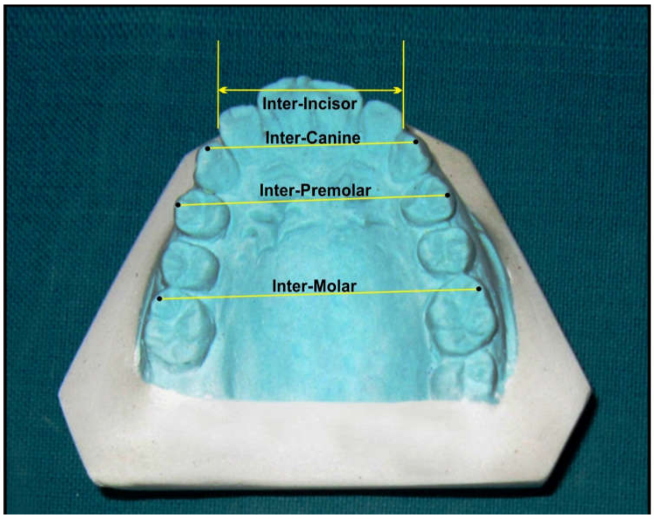

Fig1a Maxillary arch width measurements

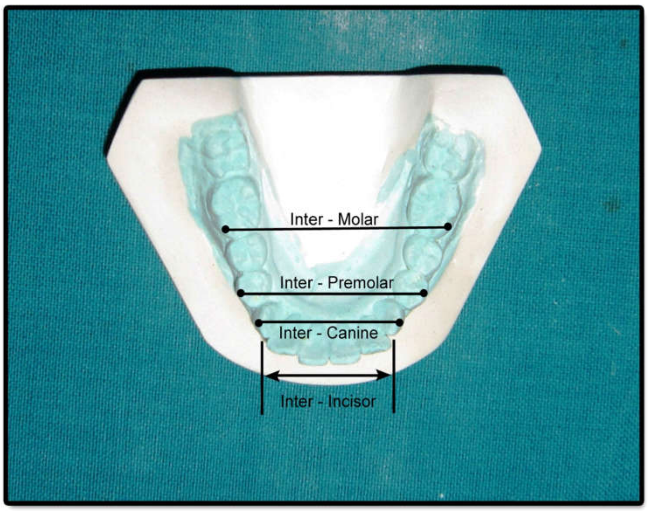

Fig 1b Mandibular arch width measurements

These measurements were taken from the distal contact points of right and left lateral incisor, the canine tips, and the cusp tips of the first premolars, molars, and mesiobuccal cusp tips of the first molars. (Fig 2a and 2b) Arch depth was measured as the shortest distance connecting the distal surface of the first permanent molar to the labial surface of the most anterior tooth in the arch.(Fig 3a and 3b)

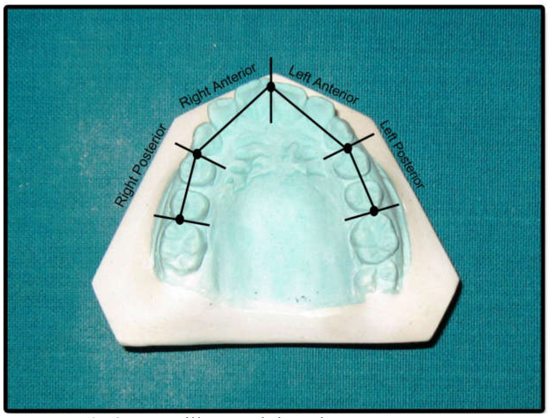

Fig 2a Maxillary arch length measurements

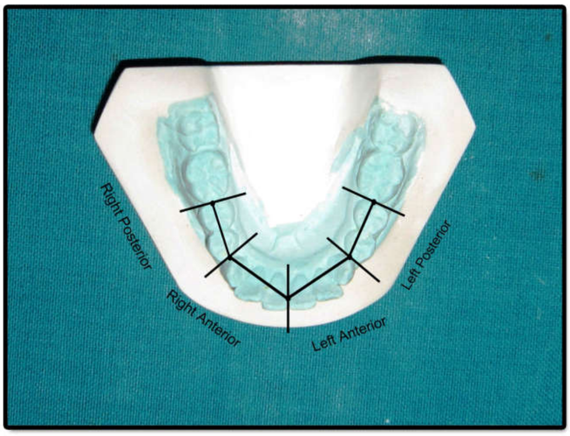

Fig 2b Mandibular arch length measurements

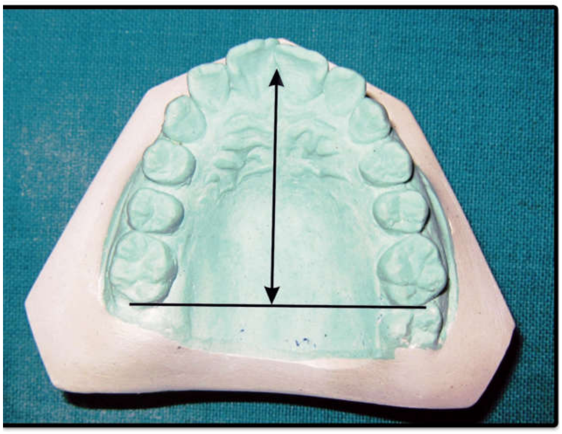

Fig 3a Maxillary Arch depth measurements

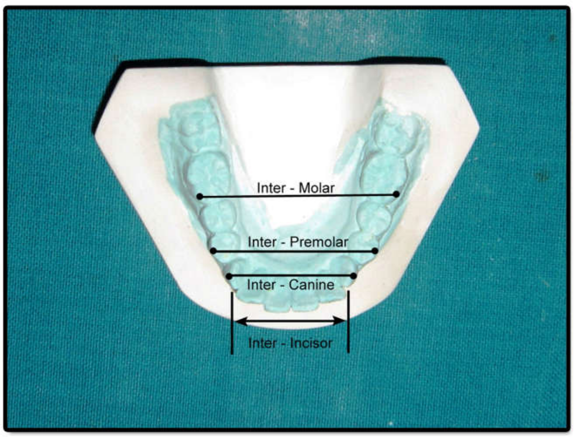

Fig 3b Mandibular arch depth measurements

Statistical analysis

For error estimation, the data on maxillary and mandibular parameters were obtained on selected control and thalassemic subjects. Error estimation was done by Dahlberg's formula**.** Table 1 provides the measurement error quantified using Dahlberg's error formula. Its expression is given by

$$D = \sqrt{\sum_{i=1}^{N} \frac{d_i^2}{2N}}$$

where D is the Dahlberg's estimated error, d_i is the difference between first and second measurement and N is the sample size which was re-measured. This estimation of standard deviation of measurement error provides the root of mean square error. It shows a maximum error measurement for Inter-incisor width of maxillary arch (0.1973) in control group, followed by inter-canine width of mandibular arch (0.1501) the same group. All other error values were less than 0.1. In TM group, all the parameters had error values less than 0.1, except inter-incisor width of mandibular arch (0.1071). The difference in the means of length, width and depth of maxillary arch in control and TM groups was evaluated for statistical significance using t-test for independent samples. The complete analysis was performed using SPSS 11.0 (IBM Inc., Armonk, NY) software package and the level of significance was 5%

Table 1 Quantification of measurement error in control and thalassemia major groups

Maxillary arch width dimension (mm)	Dahlberg's error		Mandibular arch width dimension (mm)	Dahlberg's error	
	Control group	TM group		Control group	TM group
Inter-incisor	0.1973	0.0328	Inter-incisor	0.0806	0.1071
Inter-canine	0.1022	0.097	Inter-canine	0.1501	0.0168
Inter- premolar	0.0314	0.0592	Inte- premolar	0.036	0.008
Inter-molar	0.0761	0.0163	Inter-molar	0.024	0.0136
Length in mm			Length in mm		
Left anterior	0.0983	0.0159	Left anterior	0.0263	0.0143
Right anterior	0.0328	0.0127	Right anterior	0.011	0.0032
Left posterior	0.0719	0.0133	Left posterior	0.0145	0.0372
Right posterior	0.023	0.0545	Right posterior	0.0747	0.0179
Depth	0.0368	0.021	Depth	0.038	0.0202

RESULTS

As shown in Table 2, significant difference (P=0.038) was found in the mean inter-incisor width of control group (28.21 ± 1.65 mm) and TM group (27.34 ± 2.41 mm). Further, the mean inter-molar width in control group (52.64 ± 3.13 mm) significantly differed than the mean of TM group (50.92 ± 2.73 mm) having *P value* 0.0043. Statistically insignificant differences were observed for mean inter-canine width and inter-premolar width (P > 0.05). The length measurements obtained for anterior and posterior arches showed statistically insignificant difference of means between control and TM groups (P> 0.05). The depth of maxillary arch in control group (17.26 ± 2.37 mm) was significantly lower than that of the TM group (19.79 ± 4.76) having P< 0.05.

Similarly, the mean width and length of various mandibular parameters were compared between control and TM groups. Table 3 shows that the mean inter-incisor width of mandibular arch in control group (20.43 ± 2.29 mm) was significantly smaller than the mean of TM group (21.85 ± 2.45 mm) with a P value of 0.0036 at 5% level of significance. Further, the mean inter-canine width in control group (28.71 ± 2.45 mm) was significantly smaller than that of the TM group (29.94 ± 3.40 mm) having *P* value 0.0404. The mean inter-premolar width in control group (33.99 ± 2.76 mm) differed very significantly (*P*< 0.0001) from that of TM group (36.80 ± 2.54 mm). However, the mean inter-molar width did not differ significantly between two groups.

Table 2 Mean and standard deviation of width, length, perimeter and depth of maxillary arch in control and thalassemia major groups

Maxillary arch	Study groups		
	Control group (n=50) Mean ± SD	TM group (n=50) Mean ± SD	p-value
Width (mm)			
Inter-incisor	28.21 ± 1.65	27.34 ± 2.41	0.0380*
Inter-canine	35.69 ± 2.47	35.52 ± 2.82	0.7463†
Inter- premolar	41.09 ± 2.66	41.49 ± 4.02	0.5596†
Inter-molar	52.64 ± 3.13	50.92 ± 2.73	0.0043*
Length (mm)			
Left anterior	21.11 ± 1.63	21.44 ± 1.8	0.3452†
Right anterior	21.32 ± 1.55	21.63 ± 1.46	0.3076†
Left posterior	14.78 ± 1.21	14.55 ± 1.36	0.3641†
Right posterior	14.8 ± 1.2	14.67 ± 1.63	0.6394†
Perimeter (mm)	72.02 ± 3.89	72.29 ± 2.95	0.7013†
Depth (mm)	17.26 ± 2.37	19.79 ± 4.76	0.0013*

*Statistically significant at 5% level; †Not significant

Table 3 Mean and standard deviation of width, length, perimeter and depth of mandibular arch in Control and Thalassemia major groups

Mandibular arch	Study groups		
	Control (n=50) Mean ± SD	TM (n=50) Mean ± SD	p-value
Width (mm)			
Inter-incisor	20.43 ± 2.29	21.85 ± 2.45	0.0036*
Inter-canine	28.71 ± 2.45	29.94 ± 3.4	0.0404*
Inter-premolar	33.99 ± 2.76	36.8 ± 2.54	0.0001*
Inter-molar	45.95 ± 5.12	44.18 ± 6.92	0.1481†
Length (mm)			
Left anterior	16.13 ± 1.68	16.73 ± 1.12	0.0408*
Right anterior	15.96 ± 1.48	16.78 ± 1.01	0.0253*
Left posterior	16.04 ± 1.89	15.42 ± 2.12	0.1276†
Right posterior	16.05 ± 1.86	15.35 ± 2.12	0.0846†
Perimeter (mm)	64.17 ± 4.55	64.89 ± 6.17	0.5112†
Depth (mm)	12.85 ± 1.96	13.61 ± 5.74	0.3781†

*Statistically significant at 5% level; †Not significant

In view of length, the mean left anterior length of mandibular arch in control group (16.13 ± 1.68 mm) showed statistically significant difference (*P*=0.0408) with that of TM group (16.73 ± 1.12 mm). Also, the right anterior length in control group (15.96 ± 1.48 mm) showed statistically significant difference (*P* =0.0253) than that of TM group (16.78 ± 1.01 mm). However, the mean posterior lengths did not differ significantly between two groups.

The mean perimeter and mean depth also showed statistically insignificant difference of means between two groups. Overall, the null hypothesis for the present study was partially rejected.

DISCUSSION

Thalassemia is the most challenging and controversial hematologic disorders affecting almost all racial groups. The homozygous form, thalassemia major exhibits severe clinical symptoms with marked orofacial defects.[1] Skeletal and craniofacial deformities are the common manifestations. Patients experience severe consequences when their erythropoetic system attempts to compensate for ineffective erythropoiesis and anemia. These compensatory efforts may be associated with massive erythroid marrow hypertrophy in medullary and extra medullary sites. Expansion of the facial bones and obliteration of the maxillary sinuses result in protrusion of the upper jaw and malocclusion. These changes may result in cosmetic abnormalities that can cause emotional distress.[13] It has also been reported that transfusion therapy may diminish or prevent development of bony abnormalities in growing patients.[14] The major oral change in thalassaemic patients is enlargement of the maxilla resulting in a "chipmunk facies".[11] Concern for physical attractiveness begins early in the childhood and low self-esteem can be found as early as 7 years of age. Thalassemic patients show major changes in dentofacial skeleton and are reflected as typical facial features in these individuals that may cause aesthetic concern and may lead them for the orthodontic treatment. An initial evaluation of adaptive longitudinal changes in the occlusion can be helpful in the orthodontic treatment especially in growing patients. Growth and development period is influenced by environmental factors, nutrition, systemic diseases and health, ethnic and individual variations.[6] Changes in the bony structure in thalassemic patients are attributed to marrow hyperplasia. Affected patients usually suffer from spacing of the teeth and forward drift of the maxillary incisors for which orthodontic treatment may be indicated.

The present study evaluated and compared dental arch dimension in thalassemic and unaffected normal children in an age group of 7-14 years. Dental arch form was studied by measuring maxillary and mandibular arch width, arch perimeter and arch depth in thalassemic patients and compared to their normal healthy peers. The course of the disease in childhood depends on maintenance of child on adequate blood transfusion because with adequate transfusion children grow and develop normally and have no abnormal physical signs.[13, 14] An inadequately transfused child develops the typical features of Cooley's anaemia with stunted growth, bossing of the skull and overgrowth of the maxillary region resulting in "mongoloid" appearance.[15] Children who have grown and developed normally throughout the first 10 years of life begin to develop symptoms of iron loading at puberty. The first indication of iron loading is usually the absence of the pubertal growth spurt and a failure of the menarche.[16]

A particular age group of 7-14 years was considered that consisted of both mixed and permanent dentition for measuring and comparing the dental arch form in different parameters. The mixed dentition stage of development corresponds to the time when early orthodontic therapy might be started in patients with severe malocclusion while permanent dentition corresponds to the time when full conventional orthodontic therapy can be started and thus the preferred age group is considered to be the favourable age for orthodontic intervention.

Various methods have been used to compare the dental arch forms of 2 groups of subjects. Photographic digitization method was used for comparing dental arch form by Burris and Harris[17] in American blacks and whites. Nojima *et. al.*[18] used the same method of photographic digitization in Caucasian and Japanese population but it did not completely separate size and shape differences. Euclidean distance matrix analysis (EDMA) is another method which provides good measurements of differences in dental arch form and supplies information about major variations by analysing the dental arch form. Ferrario *et al*. studied dental arch form using EDMA for dental arch asymmetry, sexual dimorphism and maxillary versus mandibular arch form differences in healthy subjects with sound dentitions.[19,20] However, this method is very tedious and the interpretation is difficult due to the large matrices of the form of dental arches.

In the present study, conventional linear measurement method[11] was used for different measurements to study the dental arch form in terms of arch width, arch length and arch depth on total of 100 cases. Sagittal and transverse measurements of the maxillary and mandibular dental arches were performed on the casts. According to Bishara for all practical clinical purposes, the arch width dimensions are established in the mixed dentition by 8 years of age and increase minimally until the early permanent dentition by 13 year. As most of the orthodontic treatments occur between 8 and 13 years, it is important for the clinician to take into consideration the minimal overall changes occurring in the mandibular anterior dental arch width, with time. Such changes have significant clinical implications because they may allow for a greater differential increase in the maxillary arch width during treatment.[21]

In the present investigation, significant differences were observed in the maxillary arch width and depth of the TM group which may be attributed to the altered growth due to systemic effects of TM in affected children. Because of the systemic effects of the disease, growth is retarded but on account of the compensatory erythropoiesis severe expansion is noticed in maxilla due to cancellous nature of the maxillary bone. [2,22] For mandibular arch, the mean inter-incisor width of mandibular arch in control group (20.43 ± 2.29) was significantly ($P<0.05$). Smaller than the mean of TM group (21.85 ± 2.45) The mean inter-canine width in controls (28.71 ± 2.45) was also significantly ($P<0.05$). Smaller than that of the TM group (29.94 ± 3.40). Highly significant ($P< 0.0001$) difference was found in the mean inter premolar width in control group (33.99 ± 2.76) and TM group (36.80 ± 2.54). These variations in the findings may be due to the retarded growth in thalassemic children due to its effect on multiple organs and systems of the body. The increased width of the mandible in the premolar region is attributed to the normal increase in the arch width with the eruption of canines and premolars along with the bony changes due to ineffective erythropoiesis.

The maxilla was found to be narrower and mandible was unaffected. These significant differences in the maxillary arch dimension of subjects with thalassemic and control groups are attributed to the concomitant growth retardation and bony hyperplasia in thalassemic subjects. Normally, maxillary alveolar process diverges with the eruption of teeth and the growth of the mandibular alveolar process is more parallel.[23] Hence, it was hypothesized that maxillary arch width would be different in thalassemic group than in control group on account of the bony hyperplasia observed in an attempt of continued haemopoiesis. However, in the present study, maxillary arch width dimensions have been found to be significantly less in thalassemic subjects as compared to normal subjects, thus, partially rejecting null hypothesis. In the present study patients with TM exhibited a narrower maxilla, a shorter maxilla and mandible and smaller incisor widths for the maxillary and mandibular arches when compared to unaffected subjects.

In the TM group, we presume that by 14 years of age, patient's diagnosis is fully established and in addition to the establishment of regular blood transfusion regimen, patients is taken under good parental and medical care. Conversely, as per cephalo-caudal growth gradient, mandibular growth takes place after 14 years of age. This could be correlated to the findings of the present study as mandible was found to be unaffected in thalassemic group.

The arch depth in thalassemic subjects was found to be greater than the control group. This is attributed to the growth retardation and smaller maxillary arch widths in thalassemic patients. The smaller intermolar, intercanine widths in the upper dental arch may result in impairment of normal nasal breathing, low tongue position or abnormal swallowing which in turn can result in deep palatal vaults in these patients. Arch length was measured as a sum total of anterior arch and posterior arch length. These measurements were taken from the reproducible reference points, distal contact points of right and left lateral incisor, the canine tips, the cusp tips of the first premolars and mesiobuccal cusp tips of the first molars. These points constitute the landmarks of the maxillary and mandibular dental arch form and define the breaking points of the arch and limit sectors on which different muscle groups have an action. The dimensions of the dental arches were determined according to sagittal and transverse measurements. In the present study, the comparison of arch perimeters measured for thalassemic and control group subjects was not statistically significant. The conventional linear measurement method used to analyse dental arch form is a simple and inexpensive method, as compared to other methods, it is easy to perform, has minimal error with precisely known range of error and do not require special equipments. However, in this study, the maxillary and mandibular arch perimeters did not show significant differences when compared with the control group which may be attributed to the fact that majority of our patients were on regular blood transfusions that had little effect on growth of these patients. The findings and application of this study may have clinical implications for orthodontic treatment of thalassemia patients in Central India.

CONCLUSIONS

Within the limits of this investigation, it can be concluded that, significant differences were observed in the maxillary arch width and depth of the TM group which may be attributed to the altered growth due to systemic effects of TM in affected children. The arch depth in thalassemic subjects was found to be greater than the control group. When compared to an unaffected control group, patients with TM had a narrower and shorter maxilla with deeper palatal vaults. The maxilla was found to be narrower and mandible was unaffected. These significant differences in the maxillary arch dimension of subjects with thalassemic and control groups are attributed to the concomitant growth retardation and bony hyperplasia in thalassemic subjects.

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