Zygomatic Hypoplasia in Patients with Non-syndromic Cleft Lip and Palate: A Case–Control Study

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Abstract

Background Patients operated for cleft deformities may have zygomatic hypoplasia secondary to impaired growth of the maxilla. This has, however, not been evaluated in the past.

Subjects and Methods This study was a prospective, case–control study. This included 32 patients, aged between 19 and 25 years, who were divided into cleft and non-cleft groups. The cleft group was further divided into unilateral clefts, bilateral clefts and isolated palatal clefts. In both groups, the zygoma was assessed and compared on both sides clinically using indirect photogrammetry and radiographically using 3D CT. The results were analyzed statistically using the unpaired *t* test.

Results There was a significant difference in the zygomatic projection on cleft and non-cleft sides in the unilateral cleft group. There was no significant difference in the zygomatic projection of both sides in the other subgroups and the control group.

Conclusion The present study shows that there is a global effect of palato-alveolar cleft repair on the midfacial skeleton. Further studies are required to correlate the impact of age, gender and the technique of palato-alveolar cleft repair with the quantum of malar hypoplasia.

Keywords Zygomatic hypoplasia · Cleft lip and palate · Palatoplasty · Midface hypoplasia

Introduction

The management of patients with cleft lip and palate has evolved greatly over the years. Despite this, the surgical management of these conditions is still associated with secondary deformities. One such classic deformity, seen after repair of cleft palate and alveolus, is midface hypoplasia, which is usually discussed in the literature in terms of maxillary retrognathism. However, the midface also consists of the zygomatic complex, of which the malar eminence is the key unit. This unit forms a syndesmosis with the maxilla at the zygomatico-maxillary suture. We hypothesize that, being in close anatomical proximity to the maxilla, the malar eminence may take up the repercussions of the scarred palato-maxillary muco-periosteum, which is currently considered the primary etiologic factor for cleft maxillary hypoplasia. Apart from this, the body of the zygoma develops from the first pharyngeal arch, which may genetically be programmed for a hypoplastic phenotype in patients with non-syndromic cleft lip and palate similar to the cleft maxillae. Therefore, it may be worth assessing the impact of the palato-alveolar cleft repair procedures on the growth of the zygoma. Zygomatic hypoplasia in cleft patients has seldom been discussed in the literature. The aim of this paper was to assess the incidence of zygomatic hypoplasia in patients operated for palato-alveolar clefts and discuss the factors that predispose to the development of zygomatic hypoplasia in these patients.

Methods

The study was a prospective, case–control pilot study, which was approved by the institutional review board, SRM University. The study group (Group A) included 17

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non-syndromic cleft patients between 19 and 25 years of age, who reported to our center for the correction of secondary cleft deformities. Informed consent was obtained from all patients for photogrammetry and investigations. This group was divided into various subgroups based on the type of cleft deformity that they had. Cleft patients who were associated with syndromes were excluded from the study. The control group (Group B) included age-matched, sex-matched healthy individuals from the same regional location who reported for the management of various dental issues. Neither the study group nor the control group had any history/ongoing orthodontic treatment. Both groups of patients underwent clinical, photographic and radiographic examination to assess the zygoma bilaterally. Occlusal relationships were recorded in all the patients, in order to correlate the zygomatic hypoplasia with the dentoalveolar maxillary hypoplasia.

Photographic Evaluation

Photographs were taken using Nikon D3200 24.2 megapixel DSLR camera with 18- to 55-mm lens and inbuilt grid pattern from a distance of 30 cms (1 foot). The patients were made to stand in normal head position (NHP) with the Frankfort's horizontal plane and interpupillary line parallel to the floor. Submental photographs were taken from the chin region with the patient's head tilted to maximum neck extension from the same distance. Facial landmarks of trichion–pogonion distance and interpupillary distance were measured and transferred onto the photographs to facilitate standardization prior to photogrammetry [1]. Indirect photogrammetry was performed in all the patients to assess the quantum of zygomatic hypoplasia.

Technique of Indirect Photogrammetric Evaluation

Indirect photogrammetry was performed similar to the methods described by Nagy et al. [1], which does not use standard facial landmarks for determining the midline. Facial height (trichion–menton distance) and interpupillary distance were used for standardization of the photographs. The bipupillary line, which was constructed by joining the inferior points of both the pupils, was taken as the horizontal reference line. A perpendicular dropped from the midpoint of this line was taken as the vertical reference line [2]. The point used for lateral reference was the zygion point, which corresponds to the lateral and the most prominent point on the malar eminence at the level of the infra orbital rim [3, 4]. The distance between the midline and the zygion point was then measured on both sides of the face (Figs. 1, 2).

CT Evaluation

A computed tomogram with 3D reconstruction was performed in both groups of patients, using Siemens Somatom 64-slice CT machine. One millimeter sections were generated that were used for 3D reconstruction. DICOM software (Digital Imaging and Communication in Medicine) was used for the volumetric assessment bilaterally. The following were the landmarks that we used for volumetric evaluation:

Superiorly Fronto-zygomatic suture

Posterior Plane passing through the beginning of the inferior orbital fissure

Medially Zygomatico-Maxillary suture

Laterally Plane passing through zygion point in the frontal plane

Inferiorly Plane passing through the inferior margin of the zygomatic buttress.

Statistical analysis was performed using SPSS software, which utilized the unpaired *t* test to determine significance. A *p* value of < 0.05 was taken as significant.

Fig. 1 Frontal photograph showing the indirect photogrammetry landmarks

Fig. 2 Submental picture showing zygomatic hypoplasia

Results

A total of 32 patients were enrolled in the study. Of these, 17 patients belonged to the study group, of which 13 were male and four were female (Group A). Fifteen patients belonged to the control group, of which 11 were male and four were female (Group B). Based on the type of cleft

deformity, patients in the study group were divided into three categories, namely those operated for unilateral cleft lip and palate (Table 1) (Group A1, 11 patients; 64%), those operated for bilateral cleft lip and palate (Table 2) (Group A2, five patients; 29%) and those operated for isolated cleft palate (Table 3) (Group A3, one patient; 5%).

Occlusal Relation

Clinical dental examination revealed that eight out of 11 patients in Group A1 (81%) had posterior crossbite in relation to the cleft segment with end-on relation of the anterior teeth. The remaining three patients had negative overjet of 3–5 mm with complete collapse and crossbite of the cleft segment posteriors (Table 1). In Group A2, all the patients had positive anterior overjet. However, three out of five patients (60%) had bilateral posterior crossbite, while the other two had Class I molar relation (Table 2). The single patient in Group A3 had normal Class I molar relations with a positive overjet (Table 3). The occlusal

Table 1 Indirect photogrammetric evaluation and occlusal relation in Group A1

Patient initials	Cleft side	IPD	Mid-IPD to zygion—right side	Mid-IPD to zygion—left side	Discrepancy between right and left sides	Anterior dental relation	Posterior occlusal relation
AM	Left CLAP	60	62	57 (left)	5	– 2 mm	Left crossbite with Class III molar relation
Sp	Left CLAP	58	56	52	4	End-on anterior relation	Left crossbite in relation to premolar segment with Class III molar relation
Na	Left CLAP	58	58	54	4	– 3 mm	Left crossbite in relation to premolar segment with Class III molar relation
She	Right CLAP	60	61	55	7	End-on anterior relation	End-on molar relation with no crossbite
Pu	LT CLAP	58	59	54	5	End-on anterior relation	End-on molar relation with no crossbite
Ga	Left CLAP	60	59	59	0	+ 2 mm	Crossbite in relation to left premolar segment and end-on molar relation of left side
MA	Right CLAP	60	56	62	6	End-on incisor relation	Crossbite right in relation to premolar segment and end-on molar relation of right side
UP	Right CLAP	62	55	61	6	– 4 mm	Bilateral crossbite with class III molar relation
AM	Right CLAP	60	57	59	2	– 3 mm	Bilateral crossbite with Class III molar relation
AG	Right CLAP	58	54	59	5	End-on anterior relation	Class I molar relation bilaterally
Bg	Left CLAP	58	58	58	0	– 4 mm	Bilateral crossbite with Class III molar relation

Table 2 Indirect photogrammetric evaluation and occlusal relation in Group A2

Patient initials	Diagnosis	IPD	Distance between mid-pupillary line and zygion right side	Distance between mid-pupillary line to zygion left side	Discrepancy between the right and left sides	Anterior occlusal relation	Posterior occlusal relation
BK	B/I	62	62	62	Equal	Positive overjet	Class I relation with no crossbite
Ved	B/L	62	57	61	4	Positive overjet	Class I molar relation with crossbite irt premolar segment
SN	Bilateral CLAP	62	61	58	3	Positive overjet	Class I molar relation with no crossbite
RV	B/I CLAP	58	56	56	Equal	Positive overjet	Class I molar relation with crossbite irt premolar segment
Pr	Bilateral	58	57	55	2	Positive overjet	Class I molar relation with crossbite irt premolar segment

Table 3 Indirect photogrammetric evaluation and occlusal relation in Group A3

SN	Incomplete palate	61	61	58	3	Positive overjet	Class I molar relation bilaterally
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Table 4 Indirect photogrammetric evaluation and occlusal relation in Group B

SP	58	61	59	Positive overjet with Class I molar relation
JD	62	59	59	Positive overjet with Class I molar relation
SP	60	59	58	Positive overjet with Class I molar relation
PO	58	57	57	Positive overjet with Class I molar relation
VV	64	58	58	Positive overjet with Class I molar relation
SS	59	60	60	Positive overjet with Class I molar relation
dk	58	62	62	Positive overjet with Class I molar relation
Dh	58	62	62	Positive overjet with Class I molar relation
RK	64	58	58	Positive overjet with Class I molar relation
SS	59	60	60	Positive overjet with Class I molar relation
VK	58	61	59	Positive overjet with Class I molar relation
JG	62	59	59	Positive overjet with Class I molar relation
JD	60	59	58	Positive overjet with Class I molar relation
GP	58	57	57	Positive overjet with Class I molar relation
Sk	64	58	58	Positive overjet with Class I molar relation

relationships of the study group reflected that some degree of midface hypoplasia was definitely present in most patients.

All the patients in Group B had positive overjet and Class I molar relation (Table 4).

Indirect Photogrammetry Results

The interpupillary distance, as measured by indirect photogrammetry, was found to be between 58 and 62 mm in

Table 5 Statistical evaluation: Comparison of interpupillary distance between the study and control groups

	Mean IPD	<i>T</i> value	<i>P</i> value
Group A	59.56	– 0.35	0.36
Group B	59.86		

both the study and control groups. This was not expected to be significantly different, as all the study cases were non-syndromic. The mean IPD in the study group was 59.56 mm and was 59.86 mm in the control group (Table 5). Since this was not statistically different ($p < 0.05$), the IPD line was used as a standard for other measurements, and the midpoint of the IPD line was used as the midpoint of the face.

The midpoint–zygion ($M-Z$ distance) distance was found to be different in different groups. In the unilateral cleft group (Group A1), nine out of 11 patients (81%) had deficiency of the $M-Z$ distance on the cleft side as compared to the normal side (Fig. 3). The mean deficit recorded was 5 mm (range 2–7 mm) (Table 1). Statistical analysis revealed a significant difference between the cleft and non-cleft sides in Group A1 ($p < 0.05$, Table 6).

In the group with bilateral clefts (Group A2), a similar deficit was noticed, with three out of five patients (60%) having asymmetry. The mean deficit was 3 mm (2–5 mm) (Table 2), and the hypoplasia was located in the right side in one patient and the left side in two patients. The single

Fig. 3 Zygomatic hypoplasia in unilateral cleft lip and palate patient

Table 6 Statistical evaluation: Comparison of midline–zygion distance on the cleft and non-cleft sides in patients with unilateral clefts

	Mean <i>M–Z</i>	<i>T</i> value	<i>P</i> value
Group A1 (cleft side)	55.55	4.66	0.000076
Group A1 (non-cleft side)	59.45		

patient with isolated cleft palate (Group A3) also had a deficit of 3 mm on the left side as compared to the right (Fig. 4, Table 3). Taken together, these results were not statistically significant ($p > 0.05$, Table 7). However, it was noted clinically that there was complete collapse of the premolar segment in three patients of Group A2, corresponding to the side of malar hypoplasia, leading to an extreme degree of crossbite.

The control group (Group B) also had minor differences between the right and left sides; however, the maximum difference was 2 mm and these results were not found to be statistically significant (Table 8).

We also compared the midline–zygion values of the cleft groups and control groups. There was a statistically significant difference in the mean value of both groups ($p < 0.05$, Table 9).

Fig. 4 Zygomatic hypoplasia in incomplete cleft palate patient

Table 7 Statistical evaluation: Comparison of midline–zygion distance on both sides in patients with bilateral clefts and isolated cleft palate

	Mean <i>M–Z</i>	<i>T</i> value	<i>P</i> value
Group A2, A3 (right)	59.67	1.41	0.095
Group A2, A3 (left)	57.67		

Table 8 Statistical evaluation: Comparison of midline–zygion distance on both sides in the control group

	Mean <i>M–Z</i>	<i>T</i> value	<i>P</i> value
Group B (right)	59.43	0.48	0.32
Group B (left)	59		

Table 9 Statistical evaluation: Comparison of midline–zygion distance between cleft patients and normal controls

	Mean <i>M–Z</i>	<i>T</i> value	<i>P</i> value
Group A1, A2 (cleft side)	57	– 2.74	0.0049
Group B	59.21		

The CT evaluation revealed that the patients in Group A1 had malar hypoplasia with a mean linear (midline–zygion as seen on basal view of 3D reconstruction) discrepancy of 5 mm (4–6 mm). This was secondary to the

Fig. 5 Frontal 3D CT showing the zygomatic hypoplasia with transverse maxillary hypoplasia

Fig. 6 Submental 3D CT image showing reduced projection of the zygomatic arch at the root with zygomatic hypoplasia of the left side

reduced projection of the malar eminence (Figs. 5, 6). The volume of the zygoma was found to be equal on both the sides (range 5436–6543 mm³). This was similar to the volume seen in the non-cleft group (5495–6345 mm³) [3]. The zygomatic arch of the affected side also showed lesser transverse projection from the root to the zygomatico-temporal suture when assessed visually, as compared to the unaffected side in all the patients.

Discussion

Facial clefting is a progressive, complex congenital malformation that presents with a multitude of problems in the stomatognathic system. Though the primary correction of cleft lip and palate is surgically completed in early childhood, the repercussions of these procedures are seen throughout the growth phase, which warrants protracted management protocols. Midface hypoplasia with mandibular prognathism is the classic facial profile in non-syndromic adult patients with cleft lip and palate. This has been attributed to the intrinsic deficiency in the growth potency of the maxilla, along with iatrogenically induced growth disturbances secondary to the repair of palato-alveolar clefts [5]. The malar eminence forms the cornerstone of facial esthetics. We have noticed that this eminence also takes the repercussive whiplash effect of retarded maxillary growth and presents as zygomatic hypoplasia. Not much has been reported on zygomatic hypoplasia in cleft patients; however, one study has reported an average of 28–30% incidence of zygomatic hypoplasia in operated non-syndromic CLAP patients [6]. To our knowledge, no studies till date have described the variability that can be associated with different types of facial clefts, and the present study is the first of its kind to assess the incidence of the zygomatic hypoplasia in adult patients with different patterns of facial clefts.

It has already been shown in the literature that the transverse maxillary hypoplasia seen in cleft patients is secondary to the arrested appositional growth and remodeling of the dentoalveolar component of the maxilla. This occurs due to the biphasic vascular insult caused by the labio-palatal cleft repair along with the loss of the osteogenic cambium layer of alveolar maxilla [7]. Alveolar bone grafting [8] also synergizes the growth retardation effect, as the buccal transposition flap used for the labial closure of cleft alveolus denudes the cambium layer in the region of the zygomatico-maxillary suture. This leads to healing by secondary intention and further retardation of the appositional growth at this suture [8]. These multiple areas of scarring and fibrosis negate the growth at the circum-maxillary sutures leading to maxillary hypoplasia and associated decreased projection of the zygoma [5].

Correlations drawn between the indirect photogrammetric findings and the dentoalveolar maxilla–mandibular relationship support the hypothesis that the zygomatic hypoplasia in non-syndromic CLAP patients is secondary to the lack of projection of the zygoma, which in turn is due to the hypoplastic transverse growth pattern of the affected maxilla. The CT findings also support this fact as the volume of the zygoma was equal bilaterally in all the

patients of the study group with reduced projection of the zygoma on the affected side.

The present study supports the hypothesis that palato-alveolar scarring is a primary cause for the reduced projection of the malar eminence of the affected side. FEM studies on the transverse maxillary expansion in operated patients with cleft lip and palate have shown that the primary palate (in an operated incomplete cleft palate) and the zygomatico-maxillary buttress (in an operated complete cleft palate) offer maximum resistance to the midfacial growth in transverse dimension [9]. These studies also validate the fact that malar hypoplasia could be a repercussion of palato-alveolar repair.

The role of interceptive orthodontics in preventing transverse maxillary and zygomatic hypoplasia needs to be analyzed. Interceptive orthodontics, particularly palatal expansion, has shown to aid in transverse expansion of the maxilla and opens the circum-maxillary sutures [10]. Therefore, it may be possible that maxillary expansion prior to alveolar bone grafting may aid in improving the projection of the malar eminence, as the zygomatico-maxillary suture would remain intact. However, at present, there is no literature evidence on the improvement in the transverse projection of the zygoma by palatal expansion. Studies have shown that the greater quantum of expansion is restricted to the para-nasal skeleton after palatal expansion [11]. This makes surgical interventions in the form of malar osteotomy or malar augmentation essential to improve the zygomatic projection, which would in turn enhance the facial esthetics.

The present study describes the global effect of palato-alveolar cleft repair on the midfacial skeleton and discusses the probable interceptive and corrective management modalities that can enhance the facial symmetry in patients with cleft lip and palate. However, we could not correlate the impact of age, gender and the technique of palato-alveolar cleft repair with the quantum of malar hypoplasia. A prospective, long-term evaluation on a large volume of patients, with the above factors in mind, is essential to

assess the impact of palato-alveolar repair procedures on the craniofacial skeleton.

Compliance with Ethical Standards

Conflict of interest All authors declare that they have no conflict of interest.

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