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CEPHALOMETRIC PARAMETERS IN CLEFT PALATE PATIENTS WITH AND WITHOUT ORONASAL FISTULA

Abdolreza Jamilian[1a*], Ludovica Nucci[2b], Mohammadamin Narimani[1c], Maziyar Farhadi[1d], Mona Farahmand[1e], Zinat Kamali[3], Alireza Darnahal[4g]

[1]Department of Orthodontics, Faculty of Dentistry, Cranio Maxillofacial Research Center, Tehran Medical Sciences, Islamic Azad University, Tehran, Iran
[2]Multidisciplinary Department of Medical-Surgical and Dental Specialties, University of Campania Luigi Vanvitelli, Naples, Italy
[3]Faculty of Nutrition and Food Technology, National Nutrition and Food Technology, Research Institute Shahid Beheshti, University of Medical Science, Tehran, Iran
[4]Department of Periodontics and Oral Medicine, School of Dentistry, University of Michigan, Ann Arbor, MI 48109 USA

[a]DDS, PhD, Professor; e-mail: info@jamilian.net; ORCIDiD: https://orcid.org/ 0000-0002-8841-0447
[b]DDS, PhD Student Orthodontic Program; e-mail: ludovica.nucci@virgilio.it; ORCIDiD: https://orcid.org/0000-0002-7174-7596
[c]DDS, PhD, Professor; e-mail: info@dr-narimani.com; ORCIDiD: https://orcid.org/0000-0001-6684-3121
[d]DDS; e-mail: maziyar.farhadi@yahoo.com; ORCIDiD: https://orcid.org/0000-0002-1912-8675
[e]DDS; e-mail: mona.farahmand87@gmail.com; ORCIDiD: https://orcid.org/0000-0002-0992-0263
[f]Deputy of Research &Technology; e-mail: Z.kamali@sbmu.ac.ir; ORCIDiD: https://orcid.org/0000-0003-1488-8971
[6]DDS, Post Doctorate Student of Periodontics; e-mail: alirezadarnahal@gmail.com; ORCIDiD: https://orcid.org/0000-0001-9629-731X

ABSTRACT

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Introduction Failure in the fusion of medial nasal processes and maxillary prominences would result in the development of a cleft lip while failure in merging the palatal shelves would result in the development of a cleft palate (CP). At present, patients with a cleft palate (CP) have often undergone corrective surgery before 1 year of age. However, an oronasal fistula (ONF) often remains in the palate after surgery.

Methodology In this historical cohort, 40 patients with CP, 6-10 years old, who had undergone corrective surgery at 1 year of age were selected by targeted sampling and assigned to two groups with ONF (N=20) and without ONF (n=20). The two groups were matched in terms of age and gender. All patients had an intraoral photograph of the maxilla and a lateral cephalogram. Twenty-three cephalometric parameters were measured prior to the orthodontic treatment and compared between the two groups using a t-test.

Results CP patients with ONF had significantly higher Y-axis, gonial angle, Go1, Go2, GoGn-SN, U1-SN, and lower lip to E line compared with those without ONF ($P<0.05$). The Jarabak index was significantly lower in the CP patients with ONF compared with those without it ($P<0.05$). The difference in other cephalometric parameters was not significant between the two groups ($P>0.05$).

Conclusion Patients with ONF had significantly greater growth in the vertical dimension compared with those without ONF. Yet, the difference in the anteroposterior dimension was not significant between the two groups.

KEYWORDS

Orthodontics and Dento-Facial Orthopedics; Oronasal Fistula (ONF); Cleft Lip; Cleft Palate; Cephalometric Parameters.

1. INTRODUCTION

Failure in the fusion of medial nasal processes and maxillary prominences would result in the development of a cleft lip while failure in merging palatal shelves would result in the development of a cleft palate (CP) [1]. At present, patients with a cleft palate (CP) often undergo corrective surgery before 1 year of age. However, a fistula often remains in the palate after surgery that causes complications for patients.

CP patients often experience restricted development of craniofacial structures. Controversy exists regarding the causes of growth limitation in these patients. Some researchers [2] have attributed it to the surgical repair while some other factors not related to the corrective surgery such as genetics (inherited trait), and adaptive changes due to the mechanical presence of the cleft. Growth limitations associated with CP may affect the growth pattern in all three planes of vertical, sagittal and transverse [3-5].

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***Corresponding author:** Prof. Abdolreza Jamilian, DDS, PhD, Department of Orthodontics, Faculty of Dentistry, Cranio Maxillofacial Research Center, Tehran Medical Sciences, Islamic Azad University, Tehran, Iran
Tel.: +98-22052228; Fax: +98-2122052228; e-mail: info@jamilian.net

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Oronasal fistula (ONF) is among the most common complications of corrective surgery in CP patients [6]. Its prevalence ranges from 4% to 35% following primary palatoplasty [7]. Nasal regurgitation and speech problems (mainly hypernasality) are the main symptoms associated with ONF. The location and size of the fistula and its causes are highly variable. Primarily, it occurs as the result of repair under tension. Postoperative infection may also be the cause [6].

The growth pattern of surgically-treated and untreated CP patients has been extensively studied [8-12]. However, information about the effect of presence/absence of ONP after surgery on the growth pattern is limited. Knowledge about the growth pattern of CP patients can significantly affect the functional and esthetic treatment outcomes [13]. Considering the effect of surgical repair of the cleft on the growth pattern of the jaw in CP patients and the gap of information about the effect of the presence/absence of ONF after surgery on the growth pattern, this study aimed to assess the cephalometric parameters of 6 to 10-year-olds with CP who had undergone corrective surgery at 1 year of age prior to orthodontic surgery in two groups with and without ONF.

2. MATERIALS AND METHODS

This historical cohort was conducted on 40 CP patients, 6-10 years of age, (mean age of 8.25 years) who had undergone corrective surgery at 1 year of age and presented to a private office in Tehran. All patients had an intraoral photograph of the maxilla and a lateral cephalogram prior to their orthodontic treatment. The photographs and cephalograms had been taken for treatment purposes not related to this study. All images were obtained in the habitual centric occlusion, with teeth in contact, lips relaxed, and head in natural position. The parents or legal guardians of all patients signed informed consent forms prior to their participation in the study. The study was approved by the ethics committee of School of Dentistry Islamic Azad University, Tehran, Iran (Number 22710201961064).

A total of 40 patients including 20 patients with ONF and 20 patients without ONF were selected and enrolled using targeted sampling. The patients were matched in terms of age and gender in the two groups and there were 11 males and 9 females in each group with a mean age of 8.4±1 years in the no fistula and 8.1±1 years in the fistula group. Figure 1 and 2 show the occlusal view of patients with and without ONF respectively. Their lateral cephalograms were traced manually and the following cephalometric parameters were measured on their lateral cephalograms:

SN-FH: The angle between the SN line and the Frankfurt plane
SNA: The angle at the intersection of SN line and NA line
SNB: The angle at the intersection of SN line and NB line
ANB: The angle at the intersection of NA line and NB line
Wits appraisal: The distance between AO line and BO line
Facial angle: The angle at the intersection of Frankfurt plane and N-Pog line
Y-axis: The angle at the intersection of SN line and N-Gn line
Gonial angle: The angle at the intersection of GO-Gn and Go-Ar lines
GO1: The angle at the intersection of Ar-Go line and Go-N line
GO2: The angle at the intersection of Go-N line and Go-Me line
GoGn-Sn: The angle at the intersection of Go-Gn and SN line
N-Me: The distance between N point and Me point (or posterior facial height)
S-Go: The distance between S point with Go point (or anterior facial height)
Jarabak index: The ratio of the anterior facial height to the posterior facial height
Inclination angle: The angle at the intersection of N' line perpendicular to the palatal plane
U1-SN: The angle of upper incisors relative to SN line
U1-NA: The distance from upper incisors to NA line
L1-mand.: The angle of lower incisors relative to the mandibular plane
L1-NB: The distance between the lower incisors and NB line
Interincisal angle: The angle between the maxillary and mandibular incisors
Nasolabial angle: The angle between the line tangent to the nasal base and the line tangent to the upper lip
Upper lip to E-line: The distance between the upper lip to the Pn-Pog' or E-line
Lower lip to E-line: The distance between the lower lip to Pn-Pog' or E-line

For higher accuracy, all the above-mentioned parameters were measured twice by the same examiner with a 2-week interval to ensure adequate intraobserver agreement. The intraclass correlation coefficient (ICC) was calculated for this purpose.

The data were analyzed using SPSS version 22 (SPSS Inc., IL, USA). The normal distribution of the data in each group was analyzed using the Kolmogorov-Smirnov test. The measured cephalometric parameters were compared between the two groups using the t-test in case of the normal distribution of the data and Mann-Whitney test in case of the non-normal distribution of the data. The level of significance was set at 0.05.

Figure 1. Occlusal view with oronasal fistula.

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Figure 4. Occlusal view without oronasal fistula.

3. RESULTS

The ICC value indicated excellent intraobserver agreement. The minimum ICC was 0.991 for the ANB while the maximum ICC was 1 for the facial angle, Wits appraisal, Inclination angle, U1-SN, L1-mand., interincisal angle, gonial angle, Jarabak index, and nasolabial angle. The mean angular measurement error was 0.5°±0.5° and the mean linear measurement error was 0.5±0.5 mm.

Table 1 presents the cephalometric parameters in the anteroposterior dimension in CP patients with and without ONF. The t-test revealed no significant difference between the two groups of CP patients with and without ONF in any of the cephalometric parameters in anteroposterior dimension (P>0.05).

Table 1. Cephalometric parameters in the anteroposterior dimension in CP patients with and without ONF.

Cephalometric parameters	*Absence of ONF*	*Presence of ONF*	*P value (t-test)*
SN-FH	8.5±2.4	10.6±3.7	0.081
SNA	78.75±3	77.5±2.5	0.23
SNB	79.45±2.5	78.25±2.3	0.12
ANB	3.5±1.9	3.4±2.2	0.9
Wits appraisal	-1.5±3.8	-2±4.8	0.72
Facial angle	80.75±3.3	78.85±7.8	0.32

Table 2 presents the cephalometric parameters in the vertical dimension in CP patients with and without ONF. The results of the t-test revealed that the Y-axis was significantly greater in CP patients with ONF compared with those without ONF (P<0.05). Also, the mean size of gonial angle was significantly larger in CP patients with ONF compared with those without it (P<0.05). Go1 and Go2 in CP patients with ONF were significantly greater than the corresponding values in patients without a fistula (P<0.05). The Go-GN-SN parameter was significantly greater in CP patients with ONF as well (P<0.05). The Jarabak index was significantly lower in CP patients with ONF compared with those without it (P<0.05).

Table 2. Cephalometric parameters in the vertical dimension in CP patients with and without ONF..

Cephalometric parameters	*Absence of ONF*	*Presence of ONF*	*P value (t-test)*
Y-axis	66.4±5.8	71.5±3.6	0.001
Gonial angle	125.3±6.9	131.9±6.4	0.003
Go1	53.2±6.3	55.55±4.8	0.04
Go2	72.15±4.9	76.85±6.2	0.012
Go-GN-SN	33.4±4.1	35.9±3.5	0.038
N-Me	113±12.6	118.7±9.6	0.116
S-Go	67.95±7.3	71.15±9.1	0.228
Jarabak Index	63.15±6.2	59.5±4.2	0.035
Inclination Angle	84.25±5	83.25±7.7	0.629

Table 3 presents the dental cephalometric parameters in CP patients with and without ONF. The U1-SN in CP patients with ONF was significantly higher than that in the group without fistula (P<0.005).

Table 3. Dental cephalometric parameters in CP patients with and without ONF.

Cephalometric parameters	*Absence of ONF*	*Presence of ONF*	*P value (t-test)*
U1-Sn	99±14.3	74.2±17.5	0.001
U1-NA	5.05±2.6	5.80± 2.8	0.445
L1-Mand	90.85±8	89.4±8.7	0.289
L1-NB	4.05±1.8	4.00±1.8	0.947
Interincisal Angle	128.05±26.5	111.63±58.2	0.962

Table 4 presents the soft tissue cephalometric parameters in CP patients with and without ONF. Among the soft tissue cephalometric parameters, the lower lip to E-line was significantly larger in CP patients with fistula (P<0.005).

Table 3. Soft tissue cephalometric parameters in CP patients with and without ONF.

Cephalometric parameters	*Absence of ONF*	*Presence of ONF*	*P value (t-test)*
Nasolabial Angle	100.45±18.3	96.2±16.6	0.446
Up-lip to E-line	-3.25±3.4	-3.5±4.7	0.678
Low-lip to E-line	-1.25±2.6	1.6±4.2	0.015

4. DISCUSSION

This study aimed to assess the cephalometric parameters prior to orthodontic treatment of 6-10-year-old CP patients who had undergone corrective surgery at 1 year of age in two groups with and without ONF. The results showed that in the anteroposterior dimension, no significant difference existed between the two groups. However, in the vertical dimension, patients with ONF had significantly greater vertical growth. This stark difference in the vertical dimension may be due to mouth breathing in children, causing a more vertical growth pattern in patients with ONF.

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Naqvi et al. [13] reported short maxillary height in unoperated patients with ONF. However, the patients had a vertical growth pattern in their study. Their results were in agreement with our findings. With regards to the anterior growth however the results were in contrast to ours, which may be due to the presence of a healthy control group in their study. Moreover, our study evaluated patients with and without ONF who had undergone corrective surgery. Patients with CP have a lower anteroposterior growth than healthy individuals [14,15].

Xu et al, [8] in their case-control study on 106 cephalograms of non-syndromic CP patients and 102 healthy controls in three groups of samples reported shortened cranial base length and reduced bony nasopharyngeal height in patients with unrepaired isolated CP. The patients also had short maxillary depth and height, and a posteriorly positioned maxilla. The width of the nasal cavity, maxilla and orbit had increased and they had shortened mandibular length and height. Adults had normal nasopharyngeal and mandibular morphology. Nonetheless, they still had shortened cranial base length and short maxillary depth and anterior height. The width of the nasal cavity, maxilla and orbit had increased in them. They concluded that the craniofacial growth and morphology in unrepaired isolated CP patients is significantly affected by non-surgical factors. The same results were reported by Ye et al [8]. The difference between our results and theirs is due to the presence of a healthy control group in the aforementioned studies and the fact that we evaluated patients who had undergone corrective surgery while they studied unoperated patients.

In contrast to our study Costello et al. [16] reported that the maxillary growth was much lower in unoperated patients while the posterior facial height was higher in operated patients. Liao and Mars [14] evaluated 39 patients with CP over 17 years of age in comparison with age- and gender-matched controls. They reported that patients with unilateral cleft lip and palate (CLP) had shorter height of the basal maxilla, shorter posterior length of the basal maxilla, and less protruded basal maxilla at the zygomatic level, compared with the control group. They found a significant correlation between the posterior height of the basal maxilla and size of the cleft maxillary segment [14]. The difference between their results and ours may be due to the fact that they only evaluated the maxilla while we assessed the cranial growth pattern.

Capelozza et al. [17] evaluated the craniofacial morphology in CLP patients using lateral cephalometry. They reported that CLP patients had a smaller and more protruded maxilla. Also, the anteroposterior facial height was much larger in CLP patients, and significant differences were noted between CLP patients and healthy controls in the mandible (body, ramus, gonial angle, and mandibular plane angle). The difference between their findings and ours in the anteroposterior dimension may be due to the fact that they had a healthy control group. Racial differences and number of lost teeth can also affect the results.

Yoshida et al. [18] evaluated the morphology of unoperated CLP patients using lateral cephalometry. They reported maxillary retrusion and a steeper mandible in CLP patients, and the tendency for maxillary retrusion and a steeper mandible had a direct correlation with age. They also reported the absence of labial proclination of incisor teeth in CLP patients. However, 3 patients with CP had labial proclination of the incisor teeth. Controversy in the results can be due to the difference in age range of the patients, the presence of a healthy control group and the fact that they evaluated unoperated patients. The sample size, age range of patients, technique of surgery, and racial and environmental characteristics may explain the controversy in the results of studies. Tooth loss and its time can also affect the results.

In general, the ONF presence is associated with a vertical growth pattern When diagnosing and treating CP patients with ONF, orthodontists should recognize the facial growth pattern in order to reduce undesirable instances of craniofacial development to the extent possible.

This study was somewhat limited by its sample size. In order to examine the efficacy of different corrective surgical techniques and their timing on the cranial growth pattern of CP patients more conclusively, similar procedures with larger group of participants are recommended.

5. CONCLUSION

CP patients with ONF experienced a significantly greater growth in the vertical dimension compared with those without ONF. However, no significant difference was noted in the anteroposterior dimension between the two groups.

AUTHOR CONTRIBUTIONS

AJ: study concept, data interpretation, writing and revising the report and final approval of the article. LN: study design, literature review. MN: administration, data interpretation, recruitment, statistical analysis. MAF: data gathering, data interpretation. MOF: critical revision and final approval of the article. ZK: statistical analysis and critical revision. AD: drafting, data interpretation, critical revision and final approval of the article.

CONFLICT OF INTEREST

There is no conflict of interest.

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Abdolreza JAMILIAN
DDS, PhD, Professor
Department of Orthodontics
Faculty of Dentistry
Cranio Maxillofacial Research Center
Islamic Azad University
Tehran, Iran

CV

Professor Abdolreza Jamilian is a researcher and specialist in the field of Orthodontics. He received his D.D.S (1991), MSc in Orthodontics (1998), and Fellowship of Orthognathic Surgery & Craniofacial Syndromes (2010) from the Shahid Beheshti University in Tehran, Iran. He obtained his membership of the European Board of Orthodontics in 2013. Now he is professor at the Islamic Azad University in Tehran. His practice is limited to orthodontics. He has lectured in several international congresses and has been a consultant for various journals. He has published over 200 original, peer reviewed research and review articles, 15 book chapters and more than 300 scientific communications. He holds 3 patents in the United States Patent and Trademark Office. Research interests: Class 3 malocclusion, Cleft lip and palate, Orthognathic surgery.

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Questions

1. What are the main symptoms associated with ONF?

❑a. Hypernasality;
❑b. Less growth in the vertical dimension;
❑c. Maxillary protrusion;
❑d. Mandibular retrusion.

2. According to this article, what is the difference between the growth patterns of patients with fistula and patients without fistula?

❑a. Greater growth in the anteroposterior dimension between the two groups;
❑b. Less growth in the maxilla in patients with fistula compared with those without ONF;
❑c. Greater growth in the vertical dimension in patient with fistula compared with those without ONF;
❑d. No significant difference was noted between the two groups.

3. According to this article, at what age did patients with cleft palate perform surgery?

❑a. At 6 years of age;
❑b. At 10 years of age;
❑c. Between 6 to 10 years of age;
❑d. At 1 year of age.

4. According to this article, what causes a recurrence of a fistula after surgery?

❑a. Genetic factors;
❑b. Patient age at surgery;
❑c. Patient gender;
❑d. Penetration.