

ORIGINAL ARTICLE

Prevalence of maxillary lateral incisor agenesis and associated skeletal characteristics in an orthodontic patient population

Dalia S. Bassiouny^a, Ahmed R. Afify^a, Hosam A. Baeshen^a, Downen Birkhed^{b*} and Khalid H. Zawawi^a

^aDepartment of Orthodontics, Faculty of Dentistry, King Abdulaziz University, Jeddah, Saudi Arabia; ^bProfessor emeritus at University of Gothenburg, Gothenburg, Sweden

ABSTRACT

Objectives: This study was aimed to investigate the prevalence of maxillary lateral incisor agenesis and associated skeletal characteristics in an orthodontic patient population.

Materials and methods: The records of the 1066 patients seeking orthodontic treatment were screened for maxillary lateral incisor agenesis (MLIA). The following data were recorded for each subject: age; gender; unilateral or bilateral agenesis of MLI and side. The lateral cephalogram of each subject with MLIA was digitally traced. The data were compared to age-matched control orthodontic patients with skeletal Class I.

Results: The prevalence of maxillary lateral incisor agenesis was 4.9% (52 patients) in which 63.5% were females. There was a significant difference between MLIA patients and controls in sagittal relationships (ANB, Wits, AB plane, angle of convexity and Co-A/Co-Gn differential analyses) $p < 0.05$.

Conclusions: Patients with maxillary lateral incisor agenesis showed a significant tendency for skeletal Class III compared with the Class I control. This could be attributed to maxillary hypoplasia/retrognathia.

ARTICLE HISTORY

Received 27 January 2016

Revised 11 April 2016

Accepted 18 May 2016

Published online 8 June 2016

KEYWORDS

Agenesis; cephalometric characteristics; Class III; congenitally missing; maxillary lateral incisor

Introduction

The congenital absence of tooth/teeth (tooth agenesis), except the third molar, is considered one of the commonest developmental anomalies encountered in dentistry and the maxillary lateral incisors are often affected.[1–7]

Two etiological theories have been suggested for missing of maxillary lateral incisors. First, disturbances during the early developmental stages and mild dysplastic expression of the ectoderm.[4,8] Second, genetic factors have been implicated to have a role in the form of genetic mutations.[4,9] Recently, the findings of Alves-Ferreira *et al.* strengthened the role of the paired box gene 9 (*PAX9*) specifically in maxillary lateral incisors development and hence its mutation causing maxillary lateral agenesis.[10] *PAX9* also plays a role in the absence of wisdom teeth.[11–13]

Cobourne reported that tooth agenesis appear to represent a complex multifactorial phenotype, which is influenced by several factors including gene function, epigenetic influences, environmental interaction and developing timing.[9]

Several studies have been directed to determine the prevalence of congenitally missing upper lateral incisors in different populations. Pinho *et al.* reported the prevalence of congenitally missing laterals to be 1.3% in a Portuguese population.[14] Thilander and Myrberg reported a higher incidence in Swedish school children ranging between 3.5 and 8.8%.[15] Ploder *et al.* showed that the prevalence of hypodontia differs amongst ethnic groups ranging from 3.4% to 10.1%.[7] In Saudi Arabia, the prevalence of congenitally missing maxillary lateral incisors was found to be 2.6%.[16]

Clinically, early diagnosis and management of lateral incisor agenesis is important to improve esthetics and to avoid possible functional complications such as malocclusion and lack of alveolar growth caused by tooth agenesis.[17] Moreover, it is essential to take care during treatment planning since such individuals could be associated with craniofacial morphological changes that could affect attaining proper occlusion.[2,3,17]

Even though the maxillary lateral incisor agenesis is considered a common developmental anomaly,[2,4,18–20] limited numbers of studies were conducted to evaluate the effects of the tooth agenesis on dento-skeletal structures.[3,4,17]

Therefore, the aim of this investigation was to study the prevalence of MLIA (unilateral and bilateral) in patients seeking orthodontic treatment and their cephalometric characteristics and to compare it to skeletal Class I controls from the same populations.

Materials and method

In this study, the pre-treatment records (case histories, panoramic and cephalometric radiographs) of patients seeking orthodontic treatment at the Department of Orthodontics, Faculty of Dentistry, King Abdulaziz University, Saudi Arabia, between 2000 and 2007, were assessed and the records of subjects with maxillary lateral incisors agenesis (MLIA), unilateral or bilateral, were collected. Subjects with developmental anomaly such as cleft lip or palate, Down's syndrome or ectodermal dysplasia, history of extraction of the permanent

dentition, history of trauma, prior orthodontic treatment or patients with hypodontia were excluded. Also, if an accurate diagnosis of the agenesis could not be made from these records, the subject was excluded from the study. The study was accepted by the Research Ethics Committee at the Faculty of Dentistry, King Abdulaziz University and was conducted in accordance to ethical principles of the Helsinki declaration.

The following data were recorded for each subject with MLIA: age; gender; unilateral or bilateral agenesis of MLI and side. The lateral cephalometric radiograph of each subject with MLI agenesis was traced using Dolphin imaging software (Chatsworth, CA, USA). The following cephalometric measurements were performed: SNA, SNB, ANB, AB plane, Wits appraisal, maxillary length (Co-A), mandibular length (Co-Gn), maxillomandibular differential, facial convexity, angle of convexity, mandibular plane angle, upper and lower incisors position and angle, nasolabial angle and upper and lower lips position to E-Line.

To evaluate skeletal characteristics of the MLIA group, 40 subjects from the same population with similar age and gender having Class I malocclusion, skeletal Class I with acceptable soft tissue profile and facial harmony were collected and served as a control group. Their skeletal characteristic was determined by the ANB angle, AB plane and Wits appraisal.

Twenty records from with and without agenesis of MLI were selected randomly and evaluated by the same investigator and another investigator two weeks after the initial survey and 100% agreement between both investigators was obtained in the identification of the MLIA pattern. To insure cephalometric measurements reproducibility, one investigator performed the digitization and cephalometric analysis. To determine the errors associated with cephalometric measurements, twenty lateral cephalometric films of subjects with and without MLIA were randomly selected and re-measured by the same investigator two weeks later. The coefficients of reliability of the measurements were between 0.93 and 0.97.^[21]

Statistical analyses

Data were tabulated and analyzed using the Statistical Package for Social Science (IBM SPSS Statistics for Mac, Version 20, Armonk, NY: IBM Corp). The results were shown as mean \pm standard deviation (SD). The Shapiro-Wilk test was used to assess for normality assumption. The results showed that the data was not normally distributed and hence, the Mann-Whitney U non-parametric test was used to compare the cephalometric variables between subjects. Comparisons between subjects with MLIA (unilateral and bilateral) and controls for age were carried out using the independent sample *t*-test. The Chi square test was used when appropriate. A value of $p < 0.05$ was considered to be significant

Results

A total of 1066 orthodontic patient's records were screened and 52 patients were diagnosed with MLIA (4.9%). There was

no significant difference between the age for MLIA (mean = 19.9 years) and the control group (mean = 21.2 years), $t_{df=90} = 0.9$, $p = 0.4$.

Forty patients had bilateral MLIA (76.9%). Demographic data and distribution of the MLIA patients is presented in Table 1.

MLIA was found more frequently in females 33/52 (63.5%) than males 19/52 (36.5%), however, the Chi square test showed that the difference in distribution was not significant, $p = 0.46$ (Table 1). Furthermore, the distribution of unilateral MLIA between right and left side was different. MLIA was more frequently observed on the right than on the left and the distribution was also different between males and females, $p = 0.01$, (Table 2).

Comparison of cephalometric analysis between the unilateral and bilateral MLIA subjects showed that there was no significant difference in all cephalometric measurements, $p > 0.05$. Also, there were no significant differences between males and females in all cephalometric measurements, $p > 0.05$, therefore, the data from both groups were pooled into one MLIA group

When comparing the cephalometric variables between MLIA group ($n = 52$) and controls ($n = 40$), a statistical significance difference was found in ten out of eighteen cephalometric measurements as shown in Table 3.

Table 1. Distribution and comparisons of patients with maxillary lateral incisor agenesis.

Gender	Unilateral MLIA	Bilateral MLIA	Total	<i>p</i> Value
Males	5 (41.7%)	14 (35%)	19 (36.5%)	0.46
Females	7 (58.3%)	26 (65%)	33 (63.5%)	
Total	12 (23.1%)	40 (76.9)	52 (100%)	

Table 2. Distribution and comparisons of patients with unilateral maxillary lateral incisor agenesis.

	Right MLIA	Left MLIA	<i>p</i> Value
Males ($n = 5$)	1 (12.5%)	4 (100%)	0.01
Females ($n = 7$)	7 (87.5%)	0 (0%)	
Total	8	4	

Table 3. Cephalometric comparisons between MLIA patients and controls (data are presented as mean (\pm SD)).

Measurements	Mean (\pm SD)		<i>p</i> Value
	MLIA ($n = 52$)	Control ($n = 40$)	
SNA°	80.4 (\pm 3.9)	82.4 (\pm 3.3)	0.04*
SNB°	80.3 (\pm 3.8)	79.6 (\pm 3.6)	0.49
ANB°	0.2 (\pm 3.0)	2.8 (\pm 0.9)	<0.001*
Wits appraisal (mm)	-2.3 (\pm 3.9)	-0.2 (\pm 1.9)	0.02*
A-B plane°	-2.3 (\pm 4.4)	-4.6 (\pm 2.5)	0.01*
Co-A (mm)	74.8 (\pm 7.2)	80.6 (\pm 6.8)	<0.001*
Co-Gn (mm)	106.9 (\pm 12.5)	106.6 (\pm 9.6)	0.32
Differential (mm)	32.2 (\pm 10.5)	25.9 (\pm 5.7)	0.002*
Mandibular plane angle	32.8 (\pm 8.1)	33.5 (\pm 5.8)	0.69
Angle of convexity	2.3 (\pm 6.1)	5.6 (\pm 3.4)	0.02
Facial angle	89.5 (\pm 4.0)	89.0 (\pm 2.5)	0.90
NLA°	113.1 (\pm 12.0)	101.0 (\pm 8.3)	<0.001*
Lower lip to E-Line (mm)	-1.0 (\pm 4.1)	1.1 (\pm 2.2)	0.003*
Upper lip to E-Line (mm)	-5.9 (\pm 4.3)	-3.1 (\pm 2.15)	0.001*
Upper incisor to NA angle	23.9 (\pm 8.0)	23.6 (\pm 5.8)	0.83
Upper incisor to NA (mm)	6.8 (\pm 3.9)	6.2 (\pm 2.9)	0.65
Lower incisor to NB angle	24.6 (\pm 6.4)	27.6 (\pm 5.3)	0.019*
Lower incisor NB (mm)	6.7 (\pm 3.5)	7.01 (\pm 2.7)	0.42

*Significant difference at <0.05 level.

Based on the sagittal relationship, the cephalometric results demonstrated that MLIA subjects had more retruded maxilla, Class III skeletal profile, less convex profile, obtuse nasolabial angle, retruded upper and lower lips and retroclined lower incisors when compared to the control group (Table 3).

Discussion

Maxillary lateral incisor agenesis (MLIA) is considered one of the most common anomalies that are encountered by orthodontists in their practice. MLIA has a higher impact on esthetics, function and occlusion. The prevalence of MLIA in this study was almost 5%, which falls in the lower end of the ranges found in previous reports (between 1 and 11%).[4,14,16,19,20,22]

Furthermore, in the current study, 63.5% of patients with MLIA were females in contrast to previous reports where a small but not significant prevalence of hypodontia in females was reported.[23–25] Nonetheless, the female to male ratio was in concert with the findings of Celikoglu *et al.*[1] Moreover, significant differences have been found in earlier studies,[26,27] which is in contrast with the current findings.

The predilection of developing skeletal Class III characteristics in subjects with MLIA is an important issue among dental practitioners particularly orthodontists. In order to find out the correlation between tendency to developing skeletal Class III and MLIA, cephalometric characteristics were performed between MLIA group and an age-matched control group from the same population and not the standard cephalometric norms. The findings showed a significantly higher prevalence of skeletal Class III in subjects with MLIA than controls as shown in the cephalometric readings related to the sagittal relationship of the maxilla and mandible. The literature is still not clear about the association between MLIA and the sagittal position of the maxilla and mandible. Wisth *et al.* found maxillary retrognathism in patients with hypodontia.[28] Later, Woodworth *et al.* studied a sample of 43 patients exhibiting bilateral congenitally missing maxillary lateral incisors and concluded that accompanied craniofacial deviations from normal included a Class III tendency.[29]

The findings of this study with regards to the increased tendency towards a Class III skeletal relationship in subjects with MLIA is in concert with the findings of Chung *et al.* who studied a sample of 1622 Korean subjects and found that, compared with Classes I or II, hypodontia was more predominant in Class III subjects.[17] Furthermore, Fekonja concluded that patients with severe hypodontia showed more propensities to a skeletal Class III relationship and an increased overbite.[4] However, both these reports did not study the association between subjects with MLIA and their cephalometric characteristics and compared their results to control norms with Skeletal Class I, which was addressed in this study.

Numerous studies have been conducted in various populations examining the prevalence, gender association and possible theories of congenitally missing maxillary lateral incisors.

However, only limited studies evaluated the occlusion and cephalometric characteristics in such populations. The current study showed that the MLIA group had retrognathic maxilla when compared to the Class I control group. This was confirmed by a decrease in the Wits appraisal in addition to the tendency towards a straight or even concave profile in some cases. Dentition showed a significantly retroclined mandibular incisors which is a common feature of compensated skeletal Class III patients. Moreover, the effect of MLIA on soft tissues showed a significant difference in position of upper and lower lips to E-Line. In MLIA cases upper and lower lips were more retruded.

With regards to the vertical relation, no significant difference was found between the MLIA group and the controls (Table 3). This finding is in agreement with the conclusions reported by Chung *et al.* when they concluded that there was no association observed in subjects with hypodontia and vertical dimension or the amount of divergence.[17] Not only that, some investigators found a significantly reduced lower face height with a counter clockwise mandibular rotation in patients with hypodontia.[18,30–32]

It has been reported that treatment protocols in skeletal Class III show better outcome and prognosis when started early. Moreover, early detection and modification of such condition reduces the psychological burden of facial and dental disharmony.[33,34] The results of current study advocate clinical screening for early identification of patients who have MLIA since they have a high predilection to develop skeletal Class III relation, hence following up these patients and offering early interventions is warranted. These interventions are mostly early growth modification by either removable or fixed appliances.

One limitation to this study is that MLIA subject were only compared to Class I. However, the main objective was to evaluate if MLIA subjects do deviate from the norms. Another limitation is that the sample of this study was selected from one centre and that could bias the findings.

Conclusions

Patients with congenitally missing maxillary lateral incisors showed a significant tendency to develop skeletal Class III. This was due to maxillary hypoplasia. Orthodontists should always keep in mind while treating patients with MLIA that the predisposition of these patients to develop skeletal Class III dictates special considerations during orthodontic diagnosis and treatment planning as well as mechanotherapy. Possible early growth modification is warranted as a preventative approach for these patients.

Disclosure statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Funding information

The study was self-funded by the authors.

References

- [1] Celikoglu M, Kamak H, Yildirim H, et al. Investigation of the maxillary lateral incisor agenesis and associated dental anomalies in an orthodontic patient population. *Med Oral Patol Oral Cir Bucal*. 2012;17:e1068–e1073.
- [2] Celikoglu M, Kazanci F, Miloglu O, et al. Frequency and characteristics of tooth agenesis among an orthodontic patient population. *Med Oral Patol Oral Cir Bucal*. 2010;15:e797–e801.
- [3] Endo T, Ozoe R, Yoshino S, et al. Hypodontia patterns and variations in craniofacial morphology in Japanese orthodontic patients. *Angle Orthod*. 2006;76:996–1003.
- [4] Fekonja A. Hypodontia in orthodontically treated children. *Eur J Orthod*. 2005;27:457–460.
- [5] Mattheeuws N, Dermaut L, Martens G. Has hypodontia increased in Caucasians during the 20th century? A meta-analysis. *Eur J Orthod*. 2004;26:99–103.
- [6] Pagan-Collazo GJ, Oliva J, Cuadrado L, et al. Prevalence of hypodontia in 10- to 14-year-olds seeking orthodontic treatment at a group of clinics in Puerto Rico. *P R Health Sci J*. 2014;33:9–13.
- [7] Polder BJ, Van't Hof MA, Van der Linden FP, et al. A meta-analysis of the prevalence of dental agenesis of permanent teeth. *Community Dent Oral Epidemiol*. 2004;32:217–226.
- [8] Aktan A, Kara I, Şener İ, et al. Radiographic study of tooth agenesis in the Turkish population. *Oral Radiol*. 2010;26:95–100.
- [9] Cobourne MT. Familial human hypodontia-is it all in the genes? *Br Dent J*. 2007;203:203–208.
- [10] Alves-Ferreira M, Pinho T, Sousa A, et al. Identification of genetic risk factors for maxillary lateral incisor agenesis. *J Dent Res*. 2014;93:452–458.
- [11] Mitsui SN, Yasue A, Masuda K, et al. Novel PAX9 mutations cause non-syndromic tooth agenesis. *J Dent Res*. 2014;93:245–249.
- [12] Nieminen P, Arte S, Tanner D, et al. Identification of a nonsense mutation in the PAX9 gene in molar oligodontia. *Eur J Hum Genet*. 2001;9:743–746.
- [13] Bianchi FJ, de Oliveira TF, Saito CB, et al. Association between polymorphism in the promoter region (G/C-915) of PAX9 gene and third molar agenesis. *J Appl Oral Sci*. 2007;15:382–386.
- [14] Pinho T, Tavares P, Maciel P, et al. Developmental absence of maxillary lateral incisors in the Portuguese population. *Eur J Orthod*. 2005;27:443–449.
- [15] Thilander B, Myrberg N. The prevalence of malocclusion in Swedish schoolchildren. *Scand J Dent Res*. 1973;81:12–21.
- [16] Afify AR, Zawawi KH. The prevalence of dental anomalies in the Western region of Saudi Arabia. *ISRN Dent*. 2012;2012:837270
- [17] Chung CJ, Han JH, Kim KH. The pattern and prevalence of hypodontia in Koreans. *Oral Dis*. 2008;14:620–625.
- [18] Endo T, Ozoe R, Kubota M, et al. A survey of hypodontia in Japanese orthodontic patients. *Am J Orthod Dentofacial Orthop*. 2006;129:29–35.
- [19] Hassan DA, Abuaffan AH, Hashim HA. Prevalence of hypodontia in a sample of Sudanese orthodontic patients. *J Orthod Sci*. 2014;3:63–67.
- [20] Silva Meza R. Radiographic assessment of congenitally missing teeth in orthodontic patients. *Int J Paediatr Dent*. 2003;13:112–116.
- [21] Houston WJ. The analysis of errors in orthodontic measurements. *Am J Orthod*. 1983;83:382–390.
- [22] Stamatiou J, Symons AL. Agenesis of the permanent lateral incisor: distribution, number and sites. *J Clin Pediatr Dent*. 1991;15:244–246.
- [23] Lavelle CL, Moore WJ. The incidence of agenesis and polygenesis in the primate dentition. *Am J Phys Anthropol*. 1973;38:671–679.
- [24] Schalk-van der Weide Y, Beemer FA, Faber JA, et al. Symptomatology of patients with oligodontia. *J Oral Rehabil*. 1994;21:247–261.
- [25] Slavkin HC. Entering the era of molecular dentistry. *J Am Dent Assoc*. 1999;130:413–417.
- [26] Bergstrom K. An orthopantomographic study of hypodontia, supernumeraries and other anomalies in school children between the ages of 8–9 years. An epidemiological study. *Swed Dent J*. 1977;1:145–157.
- [27] Brook AH. Dental anomalies of number, form and size: their prevalence in British schoolchildren. *J Int Assoc Dent Child*. 1974;5:37–53.
- [28] Wisth PJ, Thunold K, Boe OE. Frequency of hypodontia in relation to tooth size and dental arch width. *Acta Odontol Scand*. 1974;32:201–206.
- [29] Woodworth DA, Sinclair PM, Alexander RG. Bilateral congenital absence of maxillary lateral incisors: a craniofacial and dental cast analysis. *Am J Orthod*. 1985;87:280–293.
- [30] Forgie AH, Thind BS, Larmour CJ, et al. Management of hypodontia: restorative considerations. Part III. *Quintessence Int*. 2005;36:437–445.
- [31] Nodal M, Kjaer I, Solow B. Craniofacial morphology in patients with multiple congenitally missing permanent teeth. *Eur J Orthod*. 1994;16:104–109.
- [32] Sarnas KV, Rune B. The facial profile in advanced hypodontia: a mixed longitudinal study of 141 children. *Eur J Orthod*. 1983;5:133–143.
- [33] Baccetti T, Franchi L, McNamara JA, Jr. Treatment and posttreatment craniofacial changes after rapid maxillary expansion and facemask therapy. *Am J Orthod Dentofacial Orthop*. 2000;118:404–413.
- [34] Yavuz I, Halicioğlu K, Ceylan I. Face mask therapy effects in two skeletal maturation groups of female subjects with skeletal Class III malocclusions. *Angle Orthod*. 2009;79:842–848.