ASYNDROMIC SUPERNUMERARY MAXILLARY LATERAL INCISOR — CASE SERIES

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Abstract
The Aim of this study was to investigate tooth size and morphology in subjects with unilateral supernumerary maxillary lateral incisor (SMLI).

Materials & Methods: The measurements were undertaken retrospectively using orthodontics study casts from archives. To assess tooth size, the mesiodistal and buccopalatal dimensions of central incisor, lateral incisor in involved side (SMLI) and uninvolved side were measurement using digital vernier caliper. Paired T test was used to determine difference in tooth size, while chi-square tests were used to assess the association between gender side and shape with midline shift.

Results: Mean mesiodistal and buccopalatal width of central and lateral incisors in the involved side 9.24mm, 7.25mm and 7.17mm, 6.58mm respectively. Whereas, in the uninvolved side it was 9.75mm, 7.83mm and 7.58mm, 7.13mm respectively.

Discussion & Conclusion: Tooth size was reduced in involved side (SMLI) with statistical significance. Upper central incisor and lateral incisor on the involved side showed greatest reduction in size. Reduced tooth dimensions in the involved size (SMLI), suggesting a shared genetic basis, and local etiological factors.

Key Words
Supernumerary maxillary lateral incisor, hyperdontia.

INTRODUCTION:
Supernumerary teeth or Hyperdontia are the additional teeth to the normal series in dental arch. They have been reported to occur in both primary and permanent dentition. Supernumerary teeth may occur singly, in multiples, unilaterally or bilaterally, in the maxilla, in mandible or both.¹

Supernumerary teeth can be classified either by shape or location. Based on shape they are further classified into two types as Eumorphic (with similar morphology to normal teeth) and Dysmorphic (small, conical or tuberculate). Based on their location they are categorized into three types. They are mesiodens (between central incisors), paramolar (buccal/palatal to molars) and distomolars (distal to molars).²

Numerous developmental conditions and syndromes have been documented to be associated with single or multiple supernumerary teeth and as part of systemic
conditions such as cleidocranial dysplasia, Gardner’s syndrome and Cleft lip and palate. In humans, the malformation of the MLI is defined as a tooth that is morphologically abnormal in shape, such as peg laterals, conical, or canine shaped. In addition, a supplemental tooth with a crown shape similar to an MLI, but 30% smaller than the antimere has been considered as a malformation.

MLI abnormalities are frequently associated with different types of Cleft lip/palate. It has been put forth that the clefting process splits the tooth germ of the MLI into two separate teeth. In children with a unilateral cleft lip and palate, Tsai (1998) observed four different distribution pattern of the primary MLI on the cleft side. These teeth were either mesial or distal to cleft or one on each side of the cleft (one of which is supernumerary), or were there is total absence. However, in patients with isolated soft tissue cleft lip, the incidence of a supernumerary maxillary lateral incisor (SMLI) varies between 40% and 73% in the primary dentition and in the permanent dentition. In the permanent dentition, SMLI was found in 42% of cleft patients. SMLI is more frequent in the deciduous dentition than in permanent one.

The development of tooth germs and the occurrence of cleft lip/palate have a close embryological relationship in terms of timing and anatomical position, and numerous studies have reported the presence of dental anomalies in association with various forms of cleft lip, cleft palate, or both. Reduced tooth size in both the mesiodistal and buccopalatal dimensions has been reported in subjects with UCLP and BCLP. Upper lateral incisors with abnormal morphology have been reported in up to 94 per cent of subjects with a cleft lip, with or without a cleft palate, on the cleft-affected side. Abnormal morphology of the upper central incisor on the cleft-affected side has also been reported.

The occurrence of SMLI in permanent dentition without any associated systemic conditions or syndrome is a very rare phenomenon. The aim of this article is to identify such rare phenomenon and provide incidence and demographic data of such rare cases from the orthodontics archives of our institution and discuss the findings and clinical implications.

**MATERIALS AND METHODS:**

From the orthodontic departmental archives, a retrospective analysis was performed to identify cases of SMLI occurring as an isolated non-syndromic trait. Cases of both genders with erupted permanent maxillary incisors and molars, reporting unilateral SMLI in non-syndromic, non-cleft individuals during a period of January 2006 – December 2008 were identified. Patients with history of previous treatment were excluded. Patient details were retrieved from case records. Details of age, gender, Side (right/left), shape (eumorphic/dysmorphic), associated anomalies in the premaxilla, midline shift, occlusion, mesiodistal width of central incisor, lateral incisor (in side where SMLI were present, involved side and contralateral, uninvolved side) and the mesiodistal width of the SMLI were noted. In addition, a supernumerary maxillary lateral incisor with a crown shape similar to MLI, but 30% less than the antimere in the orthodontic models have been considered as a malformation and is followed in this study. Mesiodistal (MD) and Buccopalatal (BP) width was measured to the nearest 0.1 millimeters from study casts by authors using digital vernier caliper along the point of maximum dimension and an average measurement taken as the final value.

Data thus collected were entered and analyzed using Statistical Package for Social Service (SPSS) Version 16.0.0. Descriptive statistics for all the variables were presented. Chi-square test was used to assess the association between gender, side and shape with midline shift. Paired t test was used to compare the mesiodistal (MD) width of central and lateral incisors in the involved and uninvolved side. A p value of < 0.05 was considered as statistically significant.

**RESULTS**

During the 2-year period, there were 1752 numbers of cases reported to department of Orthodontics and Maxillofacial orthopedics. Of these 6 cases fulfilled the study criteria and were included for the study. The incidence was 0.34% in the given period of time.

There were 3 males and 3 females in the study. The involved side revealed equal predilection for left and right side. The mean age of presentation was 21 years with a standard deviation of 5.48 years and a range of 15 to 29 years. There were 3 cases with dysmorphic SMLI and all cases presented with associated anomalies in the premaxilla and listed in table 1. The descriptive statistics are presented as table 2. Four cases presented with midline shift to uninvolved side, one case had a diastema and another midline shift was noticed in the involved side. 3 cases presented with class I occlusion and one case exhibited class II maloclusion. 2 cases revealed subdivision maloclusion with the buccal segment exhibiting class II relationship on the uninvolved side and class I on involved side.
Table 3 discusses the midline shift and influence of gender, side and shape and no statistical significance was observed. (p = 0.368, 0.223, 0.368 respectively). Table 4 discusses the Paired t-test results that were used to compare the MD width of central and lateral incisors in the involved and uninvolved side. The mean difference in the MD width of central incisors was 0.5 and the difference was statistically significant (p= 0.041) while there was mean difference of 0.42 between MD widths of MLI of both sides, it was not statistically significant. (p = 0.093). The BP width of central incisors was 7.83 and 7.25 mm in uninvolved and involved side respectively. The mean of difference was 0.2 mm and this was statistically significant. Similarly, BP width of lateral incisors for the uninvolved side was 7.17 mm while in the involved side it was 6.58 mm with a mean difference of 0.58 mm. This was statistically significant. (p = 0.001).

The upper central and lateral incisor on the involved side (presence of SMLI) were found to be significantly smaller in both dimension that those on the uninvolved side. Graph 1.

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Figure 1

Figure 2

Figure 3

Figure 4

Figure 5

Figure 6
Table 1 Supenumerary maxillary lateral incisor and associated anomalies diagnosed clinically and with models.

<table>
<thead>
<tr>
<th>SL NO</th>
<th>AGE</th>
<th>GENDER</th>
<th>INVOLVED SIDE</th>
<th>SHAPE</th>
<th>ASSOCIATED ANOMALIES IN PREMAXILLA</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>15</td>
<td>Female</td>
<td>Left</td>
<td>Dysmorphic</td>
<td>Crowding, increased overjet &amp; labially placed 21</td>
</tr>
<tr>
<td>2</td>
<td>24</td>
<td>Male</td>
<td>Right</td>
<td>Eumorphic</td>
<td>Crowding, increased overjet &amp; labially placed 11</td>
</tr>
<tr>
<td>3</td>
<td>23</td>
<td>Female</td>
<td>Right</td>
<td>Eumorphic</td>
<td>Crowding, increased overjet &amp; Midline diastema</td>
</tr>
<tr>
<td>4</td>
<td>20</td>
<td>Male</td>
<td>Right</td>
<td>Dysmorphic</td>
<td>Crowding, increased overjet &amp; labially placed 11</td>
</tr>
<tr>
<td>5</td>
<td>15</td>
<td>Male</td>
<td>Left</td>
<td>Eumorphic</td>
<td>Crowding, increased overjet &amp; labially placed 21</td>
</tr>
<tr>
<td>6</td>
<td>23</td>
<td>Female</td>
<td>Left</td>
<td>Dysmorphic</td>
<td>Crowding, increased overjet &amp; labially placed 21</td>
</tr>
</tbody>
</table>

Table 2: Descriptive statistics of the study group

<table>
<thead>
<tr>
<th></th>
<th>Mean</th>
<th>Std. Deviation</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>21.000</td>
<td>5.47723</td>
<td>15.00</td>
<td>29.00</td>
</tr>
<tr>
<td>MD width of Central incisor in the unaffected side (in mm)</td>
<td>9.7500</td>
<td>.75829</td>
<td>9.00</td>
<td>11.00</td>
</tr>
<tr>
<td>MD width of Lateral incisor in the unaffected side (in mm)</td>
<td>7.5833</td>
<td>.66458</td>
<td>6.50</td>
<td>8.00</td>
</tr>
<tr>
<td>MD width of central incisor in the affected side (in mm)</td>
<td>9.2500</td>
<td>.75829</td>
<td>8.00</td>
<td>10.00</td>
</tr>
<tr>
<td>MD width of Lateral incisor in the affected side (in mm)</td>
<td>7.1667</td>
<td>.81650</td>
<td>6.00</td>
<td>8.00</td>
</tr>
<tr>
<td>MD width of the SMLI (in mm)</td>
<td>6.0000</td>
<td>.63246</td>
<td>5.00</td>
<td>7.00</td>
</tr>
</tbody>
</table>

Table 3: Midline shift in the study group

<table>
<thead>
<tr>
<th></th>
<th>Diastema N (%)</th>
<th>Yes, to opposite side N(%)</th>
<th>Yes, to same side N(%)</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
<td>0.368</td>
</tr>
<tr>
<td>Male</td>
<td>0 (0)</td>
<td>2 (50)</td>
<td>1 (100)</td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>1 (100)</td>
<td>2 (50)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Side</td>
<td></td>
<td></td>
<td></td>
<td>0.223</td>
</tr>
<tr>
<td>Right side</td>
<td>1 (100)</td>
<td>1 (25)</td>
<td>1 (100)</td>
<td></td>
</tr>
<tr>
<td>Left side</td>
<td>0 (0)</td>
<td>3 (75)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Shape</td>
<td></td>
<td></td>
<td></td>
<td>0.368</td>
</tr>
<tr>
<td>Eumorphic</td>
<td>1 (100)</td>
<td>2 (50)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Dysmorphic</td>
<td>0 (0)</td>
<td>2 (50)</td>
<td>1 (100)</td>
<td></td>
</tr>
</tbody>
</table>
Table 4: Comparison of mean MD and BP width of Incisors between the involved and uninvolved sides by paired t test

<table>
<thead>
<tr>
<th></th>
<th>Uninvolved side</th>
<th>Involved side</th>
<th>Mean of difference</th>
<th>95% CI of difference</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>MD width of central incisors (in mm)</td>
<td>9.75</td>
<td>9.25</td>
<td>0.5</td>
<td>0.03 - 0.96</td>
<td>0.041*</td>
</tr>
<tr>
<td>MD width of lateral incisors (in mm)</td>
<td>7.58</td>
<td>7.17</td>
<td>0.42</td>
<td>-0.1 - 0.9</td>
<td>0.093</td>
</tr>
<tr>
<td>BP width of central incisors (in mm)</td>
<td>7.83</td>
<td>7.25</td>
<td>0.20</td>
<td>0.37 - 0.8</td>
<td>0.001*</td>
</tr>
<tr>
<td>BP width of lateral incisors (in mm)</td>
<td>7.17</td>
<td>6.58</td>
<td>0.58</td>
<td>0.37 - 0.8</td>
<td>0.001*</td>
</tr>
</tbody>
</table>

*Statistically significant

Graph 1 Mesiodistal and buccopalatal width of central and lateral incisor on the involved side (presence of SMLI) and uninvolved side.

- **Uninvolved side**
- **Involved side**
DISCUSSION

The human maxillary lateral incisor (HMLI) is unique in terms of its development and anatomical positioning. It is the third most common missing teeth and when present, this tooth is documented to be having most variations in term of structure including microdontia, peg shaped and dens in dente." In addition, both deciduous and permanent HMLI are anatomically positioned in an area of that is highly susceptible to insult during organogenesis. In cleft lip/palate patient, this tooth is documented to be highly involved, exhibiting as hypoplastic/delayed or ectopic eruption pattern.12

The etiopathogenesis of supernumerary teeth (ST) are explained by numerous theories including hyperactivity of dental lamina, proliferation of dental lamina, atavism, and genetic and environmental factors. Till date no satisfactory theory has been postulated to explain occurrence of ST in all cases. With a high rate of disorder involving HMLI, the occurrence of supernumerary maxillary lateral incisor (SMLI) such as the presented cases evokes the present concepts of the development of HMLI. Literature has reports of SMLI even in non-cleft cases.19, 25

From studies in Macaques, Wei (2000)17 postulated the deciduous MLI develop from medial nasal process while, Ooe18 et al proposed that HMLI develops from medial nasal process and maxillary process. This theory was demonstrated by the fact that during palate development, maxillary process delivers essential genotypic and phenotypic messages to the future premaxilla. The 3D reconstruction study19 of sequential human embryological tooth development revealed that during early 6-8 weeks growth phase, the future site of fusion of medial nasal and maxillary process is marked by 2 outgrowths of dental epithelium, separated by a narrow groove at a latter stage in an end-to-end orientation. This point coincided with the line of fusion of medial nasal and maxillary process. This study leads to the theory of two developmental site of origin of HMLI. This study established that HMLI developed along the line of fusion of maxillary and medial nasal process. This study was used to explain the occurrence of 2 lateral incisors adjacent to the cleft region6. It was observed that in such HMLI, the distal half of teeth mesial to the cleft or the mesial part of the teeth distal to cleft were always dysmorphic in nature. This probably is a result of tissue hypoplasia (decreased in number) of that particular facial process.19

Latter reports, such as that of Tondury et al20 established that dental lamina differentiation occurred after the establishment of cleft. However, the question of multiple SMLI cannot be explained by any of these theories.

To retrace embryology, formation of dental lamina of maxilla is initiated at stage 1821 and emerges as 4 isolated structures on the yet-to-be fused facial outgrowths that unite later to form the typical horseshoe shape.22 In cleft cases, resulting from rupture of the already fused facial prominences, the rupture can damage the developing dental lamina leading to abnormality in the associated HMLI.23 Under these circumstances, the medial nasal and maxillary process fuse except the dental lamina lies separated and forms SMLI. Thus a duplicated SMLI in an otherwise intact jaw could be a hypothetical evidence of incomplete cleft arising during the last phase of fusion of processes.23, 24, 25

From the results of the present study, it would be possible that a unilateral SMLI’s presence in an otherwise full complement of teeth produces a dimensional difference in central and lateral incisors in the involved side as compared to the uninvolved side. This difference was statistically significant. This could be due to a deficiency in space during odontogenesis in the involved side, and if so, would result in dysmorphism of the incisor teeth involved, which was not the case. Furthermore for an argument of mesiodistal discrepancy arising due to an extra tooth developing, there is no plausible explanation for the reduction in buccopalatal dimension with statistical significance. Hence the effect of the SMLI is likely to be not only a direct local effect on the local developing tooth germ but on the entire alveolus (Post fusion rupture). This may point to an underlying genetic link between supernumerary lateral incisor and tooth size.

The results are supported by several studies in which abnormal upper incisor morphology was found in subjects with a cleft lip, with or without a cleft palate.12, 15, 26 Solaer et al27 in his study suggested that major local disturbances related to the formation of cleft may lead to a generally high level of development instability, which in turn may lead to a difference in tooth size between the cleft and noncleft sides. These findings are in agreement with those of Foster and Lavell et al13 who suggested that the factors responsible for producing the cleft might have an adverse effect on dental development. Olin et al28 in his study on dental anomalies in cleft lip and palate theorized that an over all insufficiency of certain tissue existing in addition to possible damage to dental tooth germ during surgical repair of the cleft defect. It is assumed that this tissue deficiency directly reflects the size of teeth after eruption. The study by Abdulla et al29 lends support to
this finding. The presence of SMLI may represent an additional clinical marker for subtle oral clefts, suggesting a common genetic background between the conditions.

From a clinical point of view the existence of SMLI may cause anatomical, functional, esthetic and physiological disturbance in the maxillary anterior region. They are liable to cause crowding, displacement of other teeth, midline shift and impactions.

The current study is the first to investigate supernumerary maxillary lateral incisor to the best of author’s knowledge and first to identify a potential link between supernumerary maxillary lateral incisor and teeth adjacent to it. Though the study lacks sufficient statistical power owing to fewer cases (due to rarity), concrete conclusions cannot be drawn based on this study alone. Multicentric results across globe may yield significant understanding of this proposed theory.

CLINICAL IMPLICATION AND CONCLUSION

1. It seems more reasonable that the potential odontogenic region for the maxillary lateral incisor comes from both the medial and maxillary processes. Any factor causing non-fusion of these two processes would result in two separate odontogenic regions and hence two maxillary lateral incisors. (Supernumerary maxillary lateral incisor).

2. In case of a post-fusion rupture, the rupture line can affect the development of dental lamina, leading to supernumerary teeth in the intact jaw.

3. The variation of supernumerary maxillary lateral incisor tooth shape, either eumorphic or dysmorphic could be due to varying degree of tissue distribution by the facial processes.

4. Patients with SMLI in an otherwise intact jaw can be at increased risk to carry some genetic predisposition for oro-facial clefts.

5. This study establishes an alteration of mesiodistal and buccopalatal dimensional variation in central and lateral incisors when a supernumerary maxillary lateral incisor is found in the same quadrant.

6. Treatment for such teeth depends on the type and position of the supernumerary tooth and on its effects or potential effect on adjacent teeth or the masticatory apparatus.

7. The management of such a supernumerary tooth should form a part of a comprehensive treatment plan and no treatment shall be considered in isolation.

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25. Lisson JA, Kjaer I (1997) Location of alveolar cleft relative to the incisive fissure. Cleft Palate Craniofac J Vol 34 No.4


