Prevalence and variations of the median maxillary labial frenum in children, adolescents, and adults in a diverse population

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The median maxillary labial frenum (MMLF) is a normal anatomic structure with inherent morphologic variations. This study sought to evaluate the prevalence of those variations in a diverse ethnic population and to educate practitioners about the prevalence of MMLF variations to prevent unnecessary biopsies.

This study included adult, adolescent, and child patients at the Louisiana State University Health Science Center School of Dentistry. Among the 284 subjects examined, frenum normale was the most common frenum classification, followed by frenum with nodule and frenum with appendix. Most nodules were found in the intermediate third of the MMLF, while appendices were mainly found in the labial third. The prevalence of an appendix was significantly higher ($P < 0.001$) in Caucasians compared to African-Americans. The prevalence of nodules was marginally higher ($P = 0.096$) in Caucasians than in African-Americans. No other statistically significant differences were found with regard to ethnicity. Additionally, nodules and appendices on the MMLF were identified in all age groups, and may become more common with increasing age.

The authors determined that variations of the MMLF are inherent and do not represent a pathologic condition, nor do they require biopsy for diagnostic purposes.

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Fig. 1. Illustrations of common frenum variations.

*Normal frenum
*Persistent tectolabial frenum
*Frenum with appendix
*Frenum with nodule
*Duplication of the frenum
*Recess of the frenum
*Bifid frenum

The median maxillary labial frenum (MMLF) is a fold of mucous membrane found on the underside of the center of the upper lip that connects to the midline of the attached gingiva between the central incisors. It adapts to any of the normal movements of the lip. The MMLF is a posteruptive remnant of the tectolabial bands. Histologically, the MMLF consists of a stratified squamous epithelium that covers highly vascular, loose fibrous connective tissue with an abundance of elastic fibers. The fibrous connective tissue fibers tend to be arranged in an anteroposterior fashion, merging loosely with the submucosal fibers of the lip. At the midline alveolar insertion, the fibers blend with the mucoperiosteum and the connective tissue underlying the median suture. According to some studies, there are no muscle fibers in the MMLF. However, others have disputed this finding. As the primary teeth erupt, the height in the alveolar structures increases normally and the attachment of the frenum moves superiorly with the maxillary alveolar crest.

As with all body parts, MMLF has demonstrated variations and anomalies. Sewerin created the classifications and nomenclature that is used to describe these variations (Fig. 1). Despite this established classification system, there is controversy regarding the origins of these variations. Some authors believe that the frenal tag is familial and is inherited as an autosomal dominant trait. Others posit that nodules and other variations of
the labial frena are due to an irregularity during the development and resorption of the tectolabial frenum.11

Many articles and studies have investigated the relationship between the MMLF and the presence or absence of a diastema between the maxillary central incisors. However, a review of the English-language literature from 1936 to 2011 reveals a dearth of clinical studies (only 3 reports were found) to determine the prevalence of variations and anomalies, including the presence of nodules and appendices, in the MMLF.9,11,12 One of the studies was conducted in an isolated ethnic population, one was comprised of Peruvian children but did not report ethnicity, and one observed adults only.9,11,12

Over the past several years, it has been observed in clinical consultations and by participation in the Louisiana State University School of Dentistry (LSUSD) Oral Pathology Biopsy Service that many dentists are not aware of the variations of frena types of MMLF, thus these variations can be misinterpreted as lesions. A survey of dentists submitted to the LSUSD Oral Pathology Biopsy Service is included as part of this study and supports this observation.

This study of MMLF variations is the first since Sewerin investigated the prevalence of variations and anomalies in the MMLF in an adult population for a 1971 study.9 It also is the first study to include a diverse patient pool, with subjects from 6 different racial/ethnic backgrounds, ranging in age from 3 to 74. This study sought to evaluate the prevalence of variations in MMLF in a diverse ethnic population, and to educate practitioners about the prevalence of variations of the MMLF, thus preventing unnecessary biopsies. It was hoped that including siblings in the study population might further clarify whether the presence of a nodule or appendix on the frenum is developmental or genetic.

Materials and methods
This prospective study was approved by the Louisiana Health Science Center Institutional Review Board. Patients who had cleft lip, congenital deformities, oro-facial syndrome, and/or had experienced operations on or traumatic injuries to the labial frenum were excluded from the study. Patients were examined by 1 of 3 calibrated dentists to ensure consistency in assessment. The exam consisted of gently distending the maxillary lip and performing a visual examination. Photographs were taken at the discretion of the examiner (Fig. 2-4), and frena were categorized according to the following classifications: frenum normale, frenum with nodule, frenum with appendix, persistent tectolabial frenum, duplication of frenum, recess of the frenum, or bifid frenum. Frenum with appendix and frenum with nodule were further classified by the location of the variation; that is, the alveolar third, intermediate third, or labial third. In addition, tooth spacing was examined and classified into 1 of 4 categories: no spacing, spacing between the central incisors only, generalized spacing, or edentulous, respectively.

The authors reviewed the pathology reports of all MMLF biopsy specimens from 1977 to 2005, so that cases of appendix frena and nodule frena that had been biopsied by the LSUSD Oral Pathology Biopsy Service could be included. All cases accepted into the study fulfilled the histologic criteria for appendix frena and nodule frena as defined by Crowley.11 The demographic data of the accepted biopsied cases from the MMLF were reviewed and recorded. Data were analyzed using descriptive statistics, exact chi-square tests, and analysis of variance (ANOVA), with Tukey’s test for post hoc analysis. SAS version 9.2 (SAS Institute Inc.) was used for all data analysis.

Results
This study examined the frena of 284 subjects, 56.7% of whom were male. Nearly half (48.94%) of the subjects were Caucasian; the other ethnicities were

<table>
<thead>
<tr>
<th>Classification</th>
<th>N</th>
<th>Mean age (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frenum normale</td>
<td>197</td>
<td>17.8 (11.4)</td>
</tr>
<tr>
<td>Frenum with nodule</td>
<td>50</td>
<td>24.3 (16.8)</td>
</tr>
<tr>
<td>Frenum with appendix</td>
<td>30</td>
<td>28.2 (10.6)</td>
</tr>
<tr>
<td>Persistent tectolabial frenum</td>
<td>4</td>
<td>9.4 (2.9)</td>
</tr>
<tr>
<td>Duplication of frenum</td>
<td>3</td>
<td>30.7 (6.1)</td>
</tr>
<tr>
<td>Bifid frenum</td>
<td>2</td>
<td>29.5 (3.5)</td>
</tr>
<tr>
<td>Recess of frenum</td>
<td>1</td>
<td>25.0</td>
</tr>
</tbody>
</table>

Table. Frenum classifications found in the present study.
African-American (38.38%), American Indian (0.70%), Asian-American (5.28%), Hispanic-American (2.11%), and patients of multiple ethnic backgrounds (4.58%). To simplify subsequent analyses, the ethnicities other than Caucasian and African-American were classified as “other.” The average age of subjects participating in the study was 20.1 years (SD ± 12.9). The youngest participant was 3 years old; the oldest was 74 years old.

The most common classification of frenum was frenum normale (found in 68.64% of patients) followed by frenum with nodule (found in 17.42% of patients) and frenum with appendix (10.45% of patients) (Table). Fifty percent of the nodules were found in the intermediate third, while 50% of appendices were found in the labial third.

There was no significant association between gender and frenum classification ($X^2 = 9.5, df = 6, P = 0.149$). However, important relationships were found between ethnicity and frenum. The prevalence of an appendix was significantly higher ($X^2 = 11.6, df = 1, P < 0.001$) in Caucasians (14.1%) compared to African-Americans (1.8%). Caucasian patients also demonstrated a higher prevalence of nodules ($X^2 = 2.9, df = 1, P = 0.096$) compared to African-Americans (21.1% compared to 12.8%). No other statistically significant differences were found in terms of ethnicity.

The chi-square test did indicate a significant relationship between diastema and frenum classification ($X^2 = 34.3, df = 18, P = 0.0115$); however, it should be noted that the chi-square test may not be valid because 75% of the cells had expected cell counts of less than 5.

ANOVA indicated a significant difference in the mean age of subjects with different frenum classifications ($F_{6,280} = 5.4, P < 0.0001$) (Table). Tukey’s test indicated that subjects who had a frenum with an appendix were significantly older than subjects with a frenum normale ($P = 0.0005$) and subjects with persistent tectolabial frenum tended to be younger than subjects with normal frenum ($P = 0.0634$). Although the sample means were very different, statistical significance was not achieved due to the small number of subjects with persistent tectolabial frenum ($n = 4$). None of the other frena classification pairs demonstrated significantly different mean ages.

It was not possible to address whether a subject was at greater risk for carrying a certain variation, since all 29 subjects with siblings had a normal frenum.

Of the 64 confirmed cases of appendix and nodular frenum from the MMLF submitted to the LSUSD Oral Pathology Biopsy Service over a 28-year period, 36 were female. In terms of race, 54 females were Caucasian, 3 were African-American, and 7 were classified as “other.” The age range of female patients at the time of biopsy was 5-81, with an average age of 44.

**Discussion**

Developmental conditions of the oral cavity that typically require no treatment include Fordyce granules, leukoedema, fissured tongue, retrocusp papilla (RCP), and the inherent morphologic variations associated with MMLF. In the authors’ experience, surprisingly little has been recorded in the English-language literature regarding the various forms and clinical parameters of MMLF. Based on the present study, variations and anomalies of the MMLF are common; more than one-third of the population had some morphologic distinction.

The results found in this study are similar to those few previously published studies. In 1971, Sewerin looked at an exclusively Danish population and found 60.2% of patients had frenum normale, compared to 68.6% in the current study.9 Crowley found frenal tags in 24% of Caucasian males and roughly 1% of African-American males.11 Sewerin’s 1971 study reported a larger percent of appendix frenuli than the present study (19.9% versus 10.5%) and fewer cases of frenum with nodule than the present study (9.1% versus 17.4%).

As in the Sewerin study, the present study did not find a statistically significant difference in frena types among males compared to females; in addition, both studies reported that nodules are most common in the intermediate third and that appendices were most likely to be found in the labial third.11 In regard to ethnicity, Crowley found 1 tissue tag in 85 African-American males. Although the author did not distinguish between nodules and appendices, the present study confirms that African-Americans report fewer cases of tissue projections.11 A short-fall in the current study is that despite the representation of many ethnicities, the present study failed to include a sufficient number of Native Americans and Asian-Americans to detect a statistically significant difference among these groups.

The present study did not find as many variations and anomalies of the MMLF in children as did a 2006 report by Diaz-Pizan et al of children ≤6 years old which found persistent tectolabial frenum in 24% of subjects, compared to less than 2% of subjects in the present study, which had a larger sample size.12 In addition, Diaz-Pizan et al reported that 12% of children had frenum with nodules, compared to 18.1% of children ≤6 years old in the present study.12 The mean ages of patients in the Diaz-Pizan et al study with persistent tectolabial frenum were very different than the present study, but statistical significance was not achieved because of the small number of subjects in the present study.12

The data from this study supports the hypothesis that the appendix and nodule of the labial frenum is developmental.9

In 1956, Crowley hypothesized, “The deepening of the (upper labial) groove takes place on either side of the midpoint and leaves a residual anterioposterior fold of the tissue untouched…and remains as the tectolabial frenum. It is believed the tissue tag is a result of an irregularity in this process of resorption.”11

The present study does not contradict this developmental theory and plausibly explains the presence of nodules and appendices on the MMLF. The tissue nodules and appendices are expected remnants that show no pathologic potential and do not need to be biopsied, which suggests that the 64 biopsies performed by the LSUSD Oral Pathology Biopsy Service could have been avoided. The large number of frena biopsied by dental professionals suggests a lack of knowledge of this normal oral structure.

An MMLF with an appendix or a nodule is similar to RCP, another developmental condition that is often biopsied unnecessarily.13 The RCP is found frequently in children, manifesting as an asymptomatic lingual mandibular gingival nodule that regresses or disappears with age.13 Lack of awareness of the morphologic variations of MMLFs and RCPS can lead to unnecessary biopsies.
Future studies should consider larger samples of very young children; in addition, larger samples of siblings and twins may shed more light on the inheritance pattern of MMLF. The present study adds additional insight into the prevalence of morphologic variants in patients of different ages and ethnicities.

Summary
MMLFs possess inherent morphologic variations; nodules and appendices are found frequently on normal frenum in all age groups and may become more common with age. There are at least 7 morphological types of MMLF (Fig.1), which vary in prevalence over a wide age range. In the present study, the most common variations were frenum with nodule and frenum with appendix. African-Americans had fewer tissue projections and significantly fewer appendices on the frenum compared to Caucasians. These variations do not represent a pathological condition and do not require biopsy for diagnostic purposes.

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