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ORIGINAL RESEARCH

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Upper lip tie: A novel classification scale with improved inter-rater reliability

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Abstract

Objectives: Upper lip tie (ULT) is a clinical condition with restrictive attachment of the superior labial frenulum (SLF), which may inhibit flanging of the lips. Objective outcome studies are lacking in part due to unreliable classification systems that rely solely upon a single attachment parameter of the SLF. This study's objectives are to describe a novel 3-point classification system for ULT and compare its inter-rater reliability to the Kotlow and Stanford systems.

Methods: Five raters used the Kotlow and Stanford systems, as well as our novel 3-point scale to score images of the SLF from 20 newborns seen at our institution between September 1, 2017 and April 1, 2018. Newborn birth weight, gestational age, and demographic data were collected from the infant's medical record. Fleiss's kappa was used to calculate inter-rater reliability for all classification systems.

Results: The parameters for our novel 3-point classification system for ULT were as follows: length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio. Our novel scale yielded the highest inter-rater reliability of 0.41, compared to 0.24 and 0.25 under the Kotlow and Stanford systems.

Conclusion: While the Kotlow and Stanford systems are based upon a single anatomical parameter, our novel 3-point classification scale uses three oral parameters that encompass anatomical points of attachment as well as the maximal length of the ULT on stretch. Our classification scheme is the first to incorporate a functional parameter of the SLF, and thereby more fully characterizes ULT.

Level of Evidence: Level 4.

KEYWORDS

Kotlow classification, novel scale, superior labial frenulum, upper lip tie

1 | INTRODUCTION

The superior labial frenulum (SLF) is the mucosal soft tissue extending from the midline maxillary gingiva into the vestibule and central upper lip. In newborns, the SLF most typically inserts at the junction between the alveolar mucosa and attached gingiva, though there is considerable variation in the morphology of the SLF.[1,2] Upper lip tie (ULT) is a clinical diagnosis that describes a low-lying attachment of the SLF with restricted lip mobility.[2] While the impact of ULT remains controversial largely due to the lack of consensus on what is normal, in addition to hindering flanging of the upper lip, ULT has been implicated in maxillary incisor diastemas, poor seal, and inefficient breastfeeding.[3] Poor seal is cited as one of the main reasons for early cessation of breastfeeding, and not breastfeeding is associated with increased incidence of neonatal infections, childhood obesity, and diabetes.[4–7]

Presently, there are two primary systems for classifying the SLF. The more commonly used Kotlow classification scale details four frenulum types (Kotlow grades I-IV) based upon the insertion point of the SLF onto the gingiva.[3] Kotlow grade I is considered least restrictive, grade II describes an SLF insertion just cranial to or between the gingival margins of the central incisors, grade III encompasses an SLF just beginning to insert into the anterior papilla, and grade IV comprises an SLF inserting into the anterior papilla or wrapping onto the palate. The Stanford classification scale also focuses on the point of attachment of the SLF, and is a 3-point classification scale with generally higher inter-rater reliability as it combines Kotlow grades II and III.[6]

Though frequently used, both classification systems possess some limitations. First, both scales are based upon a single anatomical variable of SLF attachment, which may be insufficient in characterizing ULT and predicting functional restriction in upper lip movement. Functional measures, such as the maximal length of the SLF when stretched vertically, may be more predictive of flanging mobility. As a corollary, multiple anatomical and functional parameters, including horizontal tongue mobility, vertical tongue mobility, and tongue-tip shape, are used in the Bristol Tongue Assessment Tool (BTAT) in assessing the related oral tie condition ankyloglossia.[7] This multi-parameter approach in the BTAT has been validated in prior studies, exhibiting high inter-rater reliability and correlating strongly with other ankyloglossia assessment tools, such as the Hazelbaker Assessment Tool for Lingual Frenulum Function.[8,9] A similar combined anatomical and functional approach could be applied when classifying the SLF, especially considering the poor inter-rater reliability and poor correlation with functional breastfeeding outcomes exhibited by both the Kotlow and Stanford classification scales.[2,3,6,10]

In this study, we describe our workflow in developing a novel, multi-parameter ULT classification scheme, which we call the Hopkins-Rochester Assessment of Labial Frenulum (HOP-ROC) scale. We calculate and compare the inter-rater reliability among five raters of our novel HOP-ROC scale to that of the existing Kotlow and Stanford classification scales. We also use digital measurements of the anatomical features of 20 SLFs to calculate absolute accuracy of our five raters on each of the parameters of our HOP-ROC scale.

2 | MATERIALS AND METHODS

2.1 | Study design and participants

There were 150 newborns admitted to the Johns Hopkins Hospital Newborn Nursery between September 1, 2017 and April 1, 2018 who were recruited for this study. Infants diagnosed with craniofacial anomalies were excluded. Infants admitted to the neonatal intensive care unit were also excluded. This study was approved by the Johns Hopkins Institutional Review Board (IRB00285732). All parents of the newborns recruited for this study provided informed consent. All protocols involving human subjects were carried out in accordance with the Declaration of Helsinki.

Newborn birth weight, gestational age, and demographic data were collected from the infant's medical record. A standardized protocol was followed to capture images of the SLF for all 150 newborns. Each infant's upper lip was elevated and retracted to the level of the alveolar sulcus. A standard ruler was then placed into the image frame, and multiple pictures of the infant's SLF were obtained using a high-definition digital camera. ImageJ 1.51j8 software (National Institutes of Health) was used to calibrate the measurement scale for each photograph and to adjust for slight variations in focal distance. Using this calibrated scale, absolute digital measurements for the parameters of our novel classification scale were then obtained. The 20 highest quality photographs were selected for grading, and represented a variety of ULT severities assessed by the Kotlow scale.

2.2 | Choosing the parameters for our novel classification scale

In a previous study, we described the morphologic variation of the SLF among healthy newborns.[1] In that study, we described frequency distributions for the following oral parameters: length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, length from frenulum lip attachment to vermillion border, frenulum gingival attachment thickness, frenulum labial attachment thickness, free-gingival to total-gingival length ratio, and free-lip to total-lip length ratio.

Of these seven parameters, five were selected for initial further investigation for possible inclusion in our novel HOP-ROC scale: thickness at frenulum gingival attachment, thickness at frenulum labial attachment, length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio (lip attachment ratio). This decision was made based upon clinical consensus among expert pediatric otolaryngologists at two academic medical institutions. The mean of each frequency distribution from

TABLE 1 Final 3-point classification scale with risk stratification for upper lip tie

Prototype HOP-ROC scale		
Parameter	**Value**	**Score**
Thickness at frenulum gingival attachment	≤1 mm	0
	>1 mm	1
Thickness at frenulum labial attachment	≤4 mm	0
	>4 mm	1
Length of frenulum on stretch	≥4 mm	0
	<4 mm	1
Length of alveolar edge to gingival attachment length	≥2 mm	0
	<2 mm	1
Free-lip to total-lip length ratio	≥0.75	0
	<0.75	1
3-Point HOP-ROC scale		
Parameter	**Value**	**Score**
Length of frenulum on stretch	≥4 mm	0
	<4 mm	1
Length of alveolar edge to gingival attachment length	≥2 mm	0
	<2 mm	1
Free-lip to total-lip length ratio	≥0.75	0
	<0.75	1
Final risk-stratified HOP-ROC scale		
Risk category	**Total score**	
Low	0	
Medium	1, 2	
High	3	

Abbreviation: HOP-ROC, Hopkins-Rochester Assessment of Labial Frenulum.

our previous anatomical study was used to establish threshold values for these five initial parameters (Table 1).

Five raters scored SLF images from 20 newborns using the Kotlow and Stanford classification scales, as well as the five initial parameters of our prototype HOP-ROC scale. Fleiss's kappa was used to calculate inter-rater reliabilities for the Kotlow and Stanford scales, as well as for each individual parameter of our novel prototype scale. Additionally, we compared the measurements reported by our raters against the absolute digital measurements obtained using the ImageJ 1.51j8 software, and assessed the percentage of raters who correctly categorized an image for each of the five parameters of our prototype scale. For each parameter separately, the percentage of raters who correctly categorized an image were averaged across all 20 images. The three parameters from our prototype scale that yielded the highest absolute accuracies and inter-rater reliabilities, and that were deemed by expert pediatric otolaryngologists to be the most clinically relevant for ULT were selected as final parameters for our novel 3-point HOP-ROC scale. These parameters can be visualized in

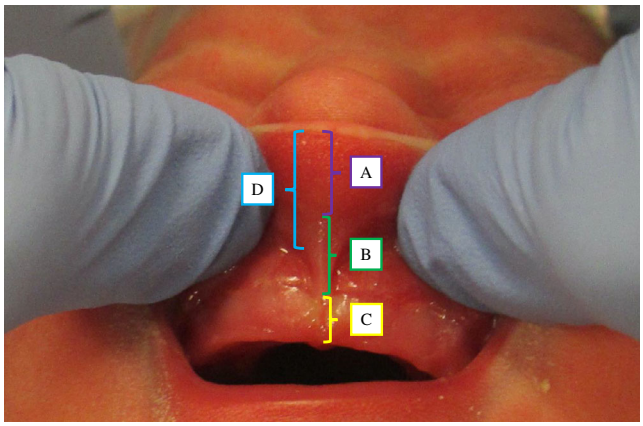

FIGURE 1 Visualization of the oral parameters for a novel upper lip tie rating scale. Anatomical parameters: A: length from frenulum labial attachment to vermillion border; B: length of frenulum on stretch; C: length from alveolar edge to frenulum gingival attachment; D: distance from sulcus to vermillion border; free-lip to total-lip length ratio is calculated as A/D.

Figure 1. We then risk-stratified our scale into the following ULT categories: 0 points as "low risk"; 1 and 2 points as "medium risk"; and 3 points as "high risk" (Table 1) to make our scale clinically informative and comprehensible for both clinicians and parents.

Inter-rater reliability from our final risk-stratified HOP-ROC scale was calculated using Fleiss's kappa, and compared to the inter-rater reliability for the Kotlow and Stanford systems. Figure 2 illustrates the overall workflow for this study.

3 | RESULTS

3.1 | Demographic information and inter-rater reliability of the five initial parameters for our prototype HOP-ROC scale

Of the 20 infants recruited, nine were male (45%). The cohort was comprised of 55% non-Hispanic White and 45% Black infants. The average (±SD) weight at birth was 3.3 ± 0.5 kg. Absolute digital measurements of the thickness at frenulum gingival attachment, thickness at frenulum labial attachment, length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio were calculated. Average (±SD) digital measurements for each parameter are shown in Table 2.

Using the prototype HOP-ROC scale in Table 1, five raters scored 20 SLF images on each of the five initial parameters. Inter-rater reliability was calculated for each parameter individually. Additionally, using the digital measurements as ground-truth standards, the percentage of raters who correctly categorized an image was calculated and averaged across all 20 images for each parameter. Thickness at frenulum gingival attachment yielded the

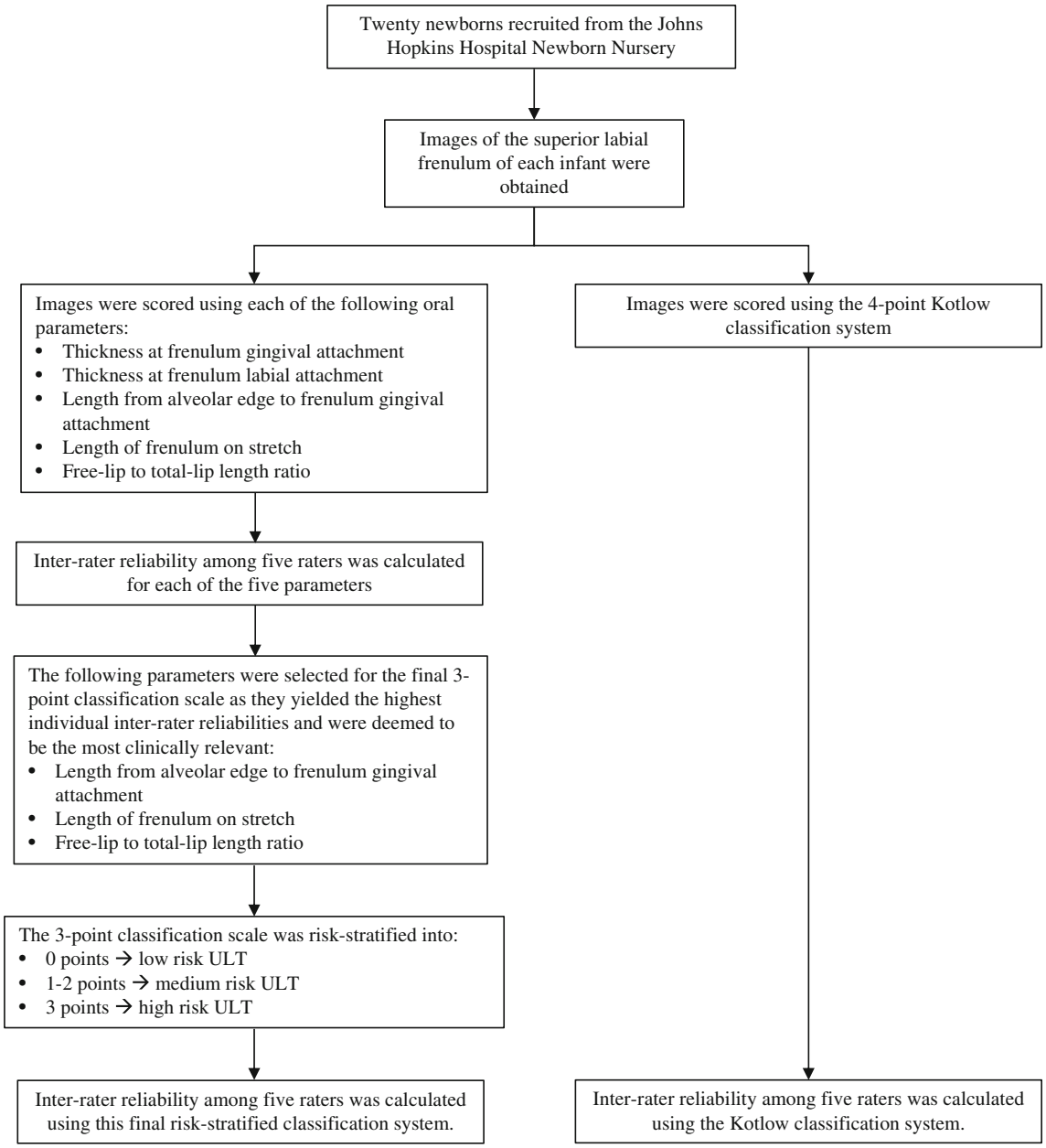

FIGURE 2 Workflow of the development of a novel 3-point classification scale for upper lip tie

TABLE 2 Measurements and inter-rater reliability of five initial oral parameters for a novel classification system for upper lip tie

Parameters	Mean (±SD), mm	Fleiss's Kappa	Average percent correct (%)
TFG	1.0 (0.6)	0.68	78
TFL	2.9 (1.2)	0.24	75
AEFG	1.6 (0.9)	0.67	78
LFS	5.6 (1.4)	0.42	87
LR	0.9 (0.1)	0.13	78

Abbreviations: AEFG, length from alveolar edge to frenulum gingival attachment; LFS, length of frenulum on stretch; LR, free-lip to total-lip length ratio; TFG, thickness at frenulum gingival attachment; TFL, thickness at frenulum labial attachment.

highest Fleiss's kappa of .68, followed by a Fleiss's kappa of .67 for length from alveolar edge to frenulum gingival attachment. Thickness at frenulum labial attachment and free-lip to total-lip length ratio yielded the lowest Fleiss's kappa coefficients of .24 and .13, respectively. All five parameters yielded greater than 75% mean absolute accuracy (Table 2).

3.2 | Inter-rater reliability of our final risk-stratified HOP-ROC classification scale

The following three parameters were selected for our final 3-point classification system based upon their inter-rater reliability coefficients, absolute accuracy scores, and expert opinions among pediatric otolaryngologists: length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio. We categorized 0 points as "low risk," 1 and 2 points as "medium risk," and 3 points as "high risk" ULT categories (Table 1).

TABLE 3 Risk categorizations of five raters using a novel, risk-stratified HOP-ROC scale for upper lip tie

	Risk categories			
Subject	Low	Medium	High	Percent agreement (%)
1	0	5	0	100
2	4	1	0	80
3	0	4	1	80
4	1	4	0	80
5	4	1	0	80
6	0	5	0	100
7	0	5	0	100
8	2	3	0	60
9	0	5	0	100
10	0	5	0	100
11	5	0	0	100
12	0	4	1	80
13	1	4	0	80
14	0	5	0	100
15	0	5	0	100
16	0	4	1	80
17	0	5	0	100
18	0	4	1	80
19	1	4	0	80
20	0	5	0	100

Abbreviation: HOP-ROC, Hopkins-Rochester Assessment of Labial Frenulum.

Categorizations and absolute percent agreement by the five raters using this novel risk-stratified HOP-ROC system are provided in Table 3 for each of the 20 SLF images. This risk-stratified classification system yielded a final Fleiss's kappa coefficient of .41, compared to a kappa coefficient of .24 and .25 using the Kotlow and Stanford systems, respectively.

4 | DISCUSSION

ULT is a clinical diagnosis characterizing a restrictive attachment of the SLF resulting in limited upper lip mobility. A comprehensive classification system that incorporates not only anatomical but also *functional* parameters, such as stretchability of the SLF, is needed to improve clinical understanding of functional outcomes and natural history of ULT. In this study, we delineate our workflow in developing a novel 3-point HOP-ROC scale for ULT which includes both anatomical and functional measures, and compare its inter-rater reliability relative to the Kotlow and Stanford systems.

The clinical impact of ULT on breastfeeding is controversial, and lack of consensus on defining ULT contributes to this controversy. This anatomic variant has been implicated in newborn feeding difficulties, as well as increased risk for central midline diastemas and dental caries due to hooding over the central maxillary incisors.[11,12] Furthermore, abnormal tethering of the upper lip may restrict superior labial movement during flanging, prevent proper latching onto the breast, and result in inefficient or painful breastfeeding.[11] This is particularly concerning given the emerging evidence suggesting a critical role of the breast milk microbiome on neonatal health.[13–16]

In developing our novel 3-point HOP-ROC scale, five oral parameters were initially investigated: thickness at frenulum gingival attachment, thickness at frenulum labial attachment, length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio. Absolute accuracy and inter-rater reliability coefficients were calculated for each individual parameter to help inform which parameters to include for our final HOP-ROC scale. Absolute accuracy for each of the five parameters was uniformly high and thereby not useful in determining our final parameters, with each parameter yielding greater than 75% average accuracy across all 20 images when assessed against ground-truth digital measurements.

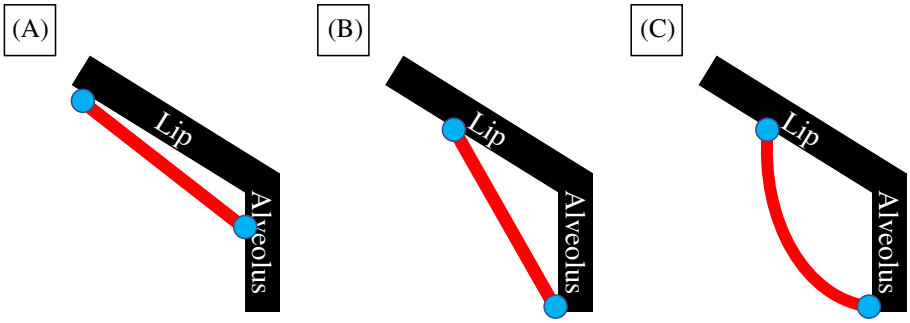

Figure legend: **blue dot**: point of attachment; **red line**: superior labial frenulum

FIGURE 3 A biomechanical model for upper lip tie from a lateral view with the upper lip lifted superiorly. Blue dot: point of attachment; red line: superior labial frenulum. Comparing A and B, though the length of the upper lip tie may be the same, different points of attachment may result in less or more restriction on upper lip movement. Comparing B and C, though the points of attachment are the same, a longer length of the lip tie may confer more range of motion of the upper lip.

Therefore, inter-rater reliability coefficients supplemented by expert opinions from pediatric otolaryngologists from two institutions were ultimately used to determine the final parameters for our novel HOP-ROC scale.

While thickness at frenulum gingival attachment yielded the highest inter-rater reliability coefficient of .68, this parameter was not chosen for our final 3-point HOP-ROC scale. This decision was based upon consensus among expert pediatric otolaryngologists that focal thickness at the site of attachment would only describe the horizontal width of the ULT at the point of attachment, and thereby yield minimal anatomical information. Functional measures regarding the mobility, flexibility, and extensibility of the lip tie, which are more likely correlated with functional breastfeeding outcomes, are better encapsulated by variables spanning the vertical axis of the ULT, though focal thickness may still be relevant for some aspects of ULT, such as a diastema.[2] For a similar reason, thickness at frenulum labial attachment was also excluded from our final scale, though this parameter also yielded a low inter-rater reliability of 0.24.

Length from alveolar edge to frenulum gingival attachment, length of frenulum on stretch, and free-lip to total-lip length ratio were the final parameters chosen for our novel 3-point HOP-ROC scale. Length from alveolar edge to frenulum gingival attachment yielded a high inter-rater reliability coefficient of .67. Anatomically, this parameter characterizes how far inferiorly the SLF attaches onto the gingiva, which is also included and is the sole anatomical parameter of the Kotlow and Stanford systems. Though length of frenulum on stretch yielded a moderate inter-rater reliability coefficient of .42, this parameter was included in our final scale as a functional measure of the flexibility and vertical extensibility of the SLF, which have been postulated in prior ULT studies as potentially impactful factors on infant breastfeeding outcomes.[2,10,17]

Free-lip to total-lip length ratio (lip attachment ratio) yielded the lowest inter-rater reliability coefficient of .13. Despite its low kappa coefficient, lip attachment ratio was included as it encapsulates the three variables that are critical to biomechanically characterize a ULT: attachment onto the upper lip, attachment onto the gingiva, and overall length of the lip tie. Figure 3 illustrates our proposed biomechanical model for ULT, and suggests why two attachment and one length variable, which are captured by lip attachment ratio, are needed to reliably reproduce a ULT. Comparing Figure 3A and Figure 3B, though the length of the ULT may be the same, different points of attachment may result in less or more restriction on upper lip movement. Likewise, a longer length of the lip tie may confer more range of motion of the upper lip compared to a shorter lip tie with the same points of attachment (Figure 3B vs. Figure 3C). The existing Kotlow and Stanford systems only take into account attachment onto the gingiva and are thereby limited in its biomechanical characterization of the ULT.

Our novel 3-point HOP-ROC system encompasses both anatomical attachments as well as flexibility and maximal length of the SLF on stretch. This scale is the first to incorporate a functional measure of flexibility and stretchability of the ULT, and thereby more fully characterizes ULT compared to existing classification schemes which rely upon a single anatomical SLF attachment parameter. Our study shows a higher inter-rater reliability of 0.41 for our novel, risk-stratified classification scheme compared to a kappa coefficient of .24 and .25 for the Kotlow and Stanford systems, which are consistent with other studies found in the literature.[2,6]

Our study was subject to several limitations. First, the practice of estimating measurements using photos may not reflect the practice of estimating measurements from patients in-person during visits. Second, further research and more widespread use are required to determine the clinical relevance of our novel classification scheme. For example, the correlation between our novel HOP-ROC scale and validated functional outcome measures, such as the LATCH score and Infant Breastfeeding Assessment Tool, should be assessed. Furthermore, functional breastfeeding outcomes likely depend not only on the infant's ULT but also upon the shape of the breast and nipple. Future studies investigating this dyad must be performed to better understand the role of a ULT on breastfeeding. We predict that the HOP-ROC scale will be more reliable for outcome studies of breastfeeding difficulties and frenotomy impact, especially since the Kotlow and Stanford systems poorly correlate with such functional outcomes.[2,10] If our novel HOP-ROC scale correlates well with functional outcomes, it may be used clinically to guide decision-making and to counsel families about the potential benefits of releasing the ULT.

5 | CONCLUSION

Our 3-point HOP-ROC system for ULT exhibits higher inter-rater reliability compared to the existing Kotlow and Stanford scales. While the Kotlow and Stanford classification schemes are each based upon a single anatomical variable, our scale incorporates three oral parameters encompassing both attachment and length of the SLF, comprehensively characterizing ULT anatomy and function/flexibility. We look forward to investigating the correlation between our scale and functional breastfeeding outcomes to further validate our classification system.

CONFLICT OF INTEREST

The authors report no conflict of interest or sources of funding.

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