

Literature Review**Management of ankyloglossia in children****Ivana Supit, Dini Widiarni Widodo**Department of Otorhinolaryngology Head and Neck Surgery, Faculty of Medicine,
Universitas Indonesia/ Dr. Cipto Mangunkusumo National General Hospital, Jakarta**ABSTRACT**

Background: Ankyloglossia, or tongue-tie, is a congenital oral anomaly characterized by an abnormally short, thick, or tight lingual frenulum that restricts tongue mobility. While mild cases may be asymptomatic, moderate to severe cases can impair essential functions such as breastfeeding, swallowing, mastication, speech articulation, and oral hygiene. In children, untreated ankyloglossia may also affect psychosocial development, including self-esteem and interpersonal communication. **Purpose:** To provide a comprehensive overview of ankyloglossia, emphasizing its anatomical and clinical aspects, classification systems, diagnostic tools, and current treatment options. **Literature review:** A narrative review was conducted using relevant literature from peer-reviewed journals and academic textbooks. The review covers various classification systems such as the Coryllos and Kotlow classifications, which assist clinicians in assessing severity, and determining management. Clinical manifestations vary with age and may include breastfeeding difficulties in infants, speech disturbances in children, and social or functional challenges in adults. Diagnostic approaches rely on clinical examination and standardized assessment tools like the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF). Management approaches include conservative methods or surgical intervention, with options such as frenotomy, frenectomy, or frenuloplasty, depending on the patient's age and symptom severity. **Conclusion:** Ankyloglossia can significantly affect oral function and quality of life. Early recognition and appropriate intervention are essential for preventing long-term complications. Standardized diagnostic criteria and evidence-based treatment guidelines are needed to optimize care and ensure consistent clinical outcomes.

Keywords: ankyloglossia, tongue-tie, frenotomy, frenectomy, frenuloplasty

ABSTRAK

Latar belakang: Ankiloglosia, atau tongue-tie, adalah kelainan kongenital pada lidah yang ditandai dengan frenulum lingual yang pendek, tebal, atau kaku, sehingga membatasi pergerakan lidah. Meskipun kasus ringan tidak menunjukkan gejala, kondisi ini dapat berdampak pada berbagai aspek kehidupan seperti gangguan menyusu pada bayi, kesulitan menelan atau mengunyah, gangguan bicara, dan higiene rongga mulut. Pada anak, jika tidak ditangani sejak dini dapat menimbulkan gangguan psikososial, seperti rasa rendah diri, atau kesulitan berkomunikasi. **Tujuan:** Untuk memberikan pemahaman tentang ankiloglosia, mulai dari struktur dan fungsi lidah, tinjauan klinis, sistem klasifikasi, metode diagnosis, serta pilihan terapinya. **Tinjauan pustaka:** Dilakukan dengan merangkum literatur dari jurnal ilmiah dan buku akademik. Beberapa sistem klasifikasi seperti Coryllos dan Kotlow digunakan untuk menilai tingkat keparahan, dan menentukan keputusan klinis. Diagnosis umumnya berbasis pemeriksaan fisik dibantu alat standar seperti Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF). Penatalaksanaan bervariasi, dari observasi pada kasus ringan, hingga tindakan pembedahan seperti frenotomi, frenektomi, atau frenuloplasti pada kasus yang mengganggu fungsi. **Kesimpulan:** Ankiloglossia merupakan kondisi yang tidak bisa diabaikan karena berpotensi mengganggu kualitas hidup sejak dini. Pengenalan dan intervensi yang tepat waktu sangat krusial untuk mencegah komplikasi jangka panjang. Diperlukan kriteria diagnosis yang lebih seragam, dan pedoman terapi berbasis bukti, agar tatalaksana ankiloglossia menjadi lebih optimal dan hasilnya konsisten.

Kata kunci: ankiloglossia, tongue-tie, frenotomi, frenektomi, frenuloplasti

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INTRODUCTION

The tongue is a muscular organ essential for various functions including speech, mastication, swallowing, and the development of oral structures. It is anchored to the mandible and hyoid bone, with its ventral surface connected to the floor of the mouth by a mucosal fold called the lingual frenulum.^{1,2} (Figure 1)

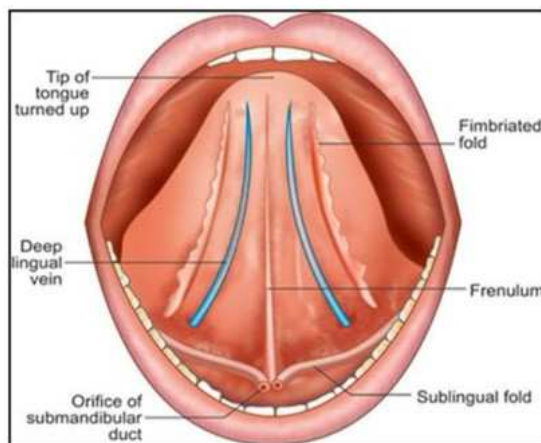


Figure 1. Anatomy of the lingual frenulum

The lingual frenulum consists of mucosal tissue and underlying connective structures that merge with the periosteum of the mandible. While it serves to stabilize the tongue, it also permits adequate mobility for normal oral function.

Tongue and frenulum development begin as early as the fourth week of gestation. Postnatally, the frenulum typically recedes or becomes less prominent as the child grows. However, in some individuals, the frenulum remains unusually short or thick, leading to restricted tongue movement.

This condition, known as ankyloglossia or tongue-tie, is a congenital anomaly characterized by reduced tongue mobility due to an abnormally tight or short lingual frenulum. Although often asymptomatic,

in more pronounced cases, especially in infants and children, it may result in significant functional impairments. These include breastfeeding difficulties, impaired articulation, challenges in chewing and swallowing, poor oral hygiene, altered jaw development, and psychosocial effects such as low self-esteem or social withdrawal.^{2,3}

Management of ankyloglossia depends on symptom severity and patient's age. While mild cases may not require intervention, moderate to severe forms often necessitate surgical treatment. Procedures such as frenotomy, frenectomy, and frenuloplasty are executed to release the frenulum and restore normal tongue function, with the choice of technique tailored to individual clinical presentation.^{4,5}

This study aimed to provide a comprehensive overview of ankyloglossia, including its anatomy, classification, epidemiology, etiopathology, clinical manifestation, diagnostic criteria, and management strategy. It further discussed the functional and psychosocial impacts of the condition, critically evaluates surgical options based on clinical presentation and severity, and highlights current challenges in diagnosis, treatment thresholds, and long-term outcomes.

LITERATURE REVIEW

Definition and classification

Ankyloglossia is a congenital condition characterized by a developmental disorder of the tongue, marked by a short and thick lingual frenulum, which cause restriction in the tongue movement. Ankyloglossia is classified into four degrees based on Kotlow's classification. Class I (mild ankyloglossia) occurs when the distance from the tip of the

tongue to the lingual frenulum is between 12 mm and 16 mm. Class II (moderate ankyloglossia) occurs when the distance is between 8 mm and 11 mm. Class III (severe ankyloglossia) occurs when the distance is between 3 mm and 7 mm. Class IV (total or complete ankyloglossia) occurs when the distance is less than 3 mm.⁴ (Table 1)

Table 1. Severity of ankyloglossia based on Kotlow's classification

Kotlow's classification	Tongue movement
Clinically accepted	>16mm
Class I mild AG	12-16mm
Class II moderate AG	18-11mm
Class III severe AG	3-7mm
Class IV complete AG	<3mm

The tongue is considered unrestricted if the distance from the tip of the tongue to the lingual frenulum exceeds 16 mm. Tongue movement is deemed normal if the tip of the tongue can be extended outside the mouth, can easily touch the upper and lower lips without being pulled, and when it is retracted, it does not show impressions of the teeth on the lingual frenulum mucosa, or cause a gap between the mandibular incisors.⁴

Epidemiology

The prevalence of ankyloglossia varies between 0.2%-5%, with males being more affected than females. Among toddlers, the incidence of ankyloglossia ranges from 25% to 60%. The prevalence in neonates is higher compared to children [(1.72%-10.7%) vs (0.1%-2.08%)]. This phenomenon is thought to occur because the lingual frenulum may thin and disappear or improve with age, or the condition may be compensated over time.

Etiopathology

Ankyloglossia may have a genetic origin or be inherited. Molecular studies in patients with ankyloglossia have identified mutations in the TBX22 gene, which is also implicated in the development of cleft lip and palate. Additionally, hereditary conditions such as epidermolysis bullosa can predispose individuals to ankyloglossia. The condition has also been linked to various syndromes, including Opitz, orofaciogigital, Beckwith-Wiedemann, Simpson-Golabi-Behmel, Van de Woude, and Pierre Robin syndrome.⁶

Clinical manifestation

Ankyloglossia is often asymptomatic and may adapt on its own, or be compensated for as the individual grows older.⁷ In children, however, ankyloglossia can lead to several challenges, including breastfeeding difficulties, speech disorder, poor oral hygiene, impaired communication, and low self-confidence.⁸

Several studies found a significant correlation between ankyloglossia and breastfeeding issues. The most common problem in infants with the condition is latching difficulty during breastfeeding. Due to limited tongue mobility, the infant struggles to create an effective seal on the nipple, relying instead on the gums to hold the nipple in place. This inefficient feeding technique reduces milk transfer, leaving the infant frustrated, and often leading to nipple biting. Consequently, the infant may suffer from malnutrition, while the mother may experience nipple trauma, pain, and mastitis caused by blocked milk ducts.⁶

Speech and articulation difficulties typically emerge as the child enters the language-learning phase. Limited tongue movement hampers clear articulation,

particularly for sounds like “R, S, Z, T, D, L, J, Zh, Ch, Th, and Dg”.⁴ Additionally, a frenulum positioned too close to the gums can exert excessive tension on the mandible, potentially causing mandibular prognathism and leading to Class III malocclusion.^{4,6} Restricted tongue mobility also hinders effective cleaning of the teeth, contributing to poor oral hygiene over time.⁶

The cumulative impact of these challenges can adversely affect a patient’s social interactions and self-esteem, further underscoring the functional and psychosocial implications of ankyloglossia.⁶

Diagnosis

The diagnosis of ankyloglossia is based on clinical and physical examination associated with the anatomical condition of the tongue. While various criteria have been proposed by clinicians to diagnose ankyloglossia, there is currently no standardized diagnostic approach. A commonly observed feature is a short frenulum, often causing the tongue to take on a ‘heart shape’ when protruded.⁸

During the evaluation, the tongue’s range of motion and functionality should be carefully assessed. Tongue movement is considered normal if the following criteria are met: (a) the tongue tip (TT) can be extended without any gap or restriction, (b) the TT can easily touch the upper and lower lips without being pulled or strained, (c) when the tongue is retracted, no bite mark appear on the mucosal surface of the lingual frenulum, (d) the lingual frenulum does not cause a gap (diastema) between the mandibular incisors, (e) infants show no signs of difficulty during breastfeeding, and (f) children exhibit no challenges in communication. Attention to these factors ensures a comprehensive assessment of tongue mobility and function in patients suspected of having ankyloglossia.⁶ (Table 2)

In addition to the criteria for the normal range of tongue motion, Hazelbaker developed another set of criteria to evaluate whether intervention is necessary for ankyloglossia. Hazelbaker assessed seven tongue movements and five tongue characteristics, as outlined in the table below.⁶ (Table 3)

Table 2. Normal range of tongue motion⁶

Tip of the tongue should protrude outside the mouth without clefting
Tip of the tongue should be able to sweep the lips easily without straining
When the tongue is retracted, it should not blanch the tissue lingual to the anterior teeth
The lingual frenulum should not create a diastema between mandibular central incisors
The lingual frenulum should not prevent the infant from attaching to the maternal nipple while nursing
Children should not exhibit speech difficulties

Table 3. Hazelbaker’s scoring questionnaire⁶

Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF)			
Appearance items	Score	Function items	Score
Appearance of tongue when lifted		Lateralization	
Round or square	2	Complete	2
Slight cleft in tip apparent	1	Body of tongue but not tongue tip	1
Heart-shaped	0	None	0
Elasticity of frenulum		Lift of tongue	
Very elastic (excellent)	2	Tip to mid-mouth	2
Moderately elastic	1	Only edges to mid-mouth	1
Little or no elasticity	0	Tip stays at alveolar ridge or rises to mid-mouth only with jaw closure	0

Length of lingual frenulum when tongue lifted		Extension of tongue	
More than 1 cm or embedded in tongue	2	Tip over lower lip	2
1 cm	1	Tip over lower gum only	1
Less than 1 cm	0	Neither of above, or anterior or mid-tongue humps	0
Attachment of lingual frenulum to tongue		Spread of anterior tongue	
Posterior to tip	2	Complete	2
At tip	1	Moderate or partial	1
Notched tip	0	Little or none	0
Attachment of lingual frenulum to inferior alveolar ridge		Cupping	
Attached to floor of mouth or well below ridge	2	Entire edge, firm cup	2
Attached just below ridge	1	Side edges only, moderate cup	1
Attached at ridge	0	Poor or no cup	0
Total appearance score:		Peristalsis	
Function items score		Complete, anterior to posterior (originates at the tip)	2
<ul style="list-style-type: none"> • 14: perfect score (regardless of <i>Appearance item score</i>) • ≥ 11: acceptable, if <i>Appearance item score</i> is 10 • < 11: function impaired 		Partial: originating posterior to tip	1
		None or reverse	0
		Snapback	
Frenotomy should be considered if management fails.		None	2
Frenotomy necessary if Appearance item score is < 8 .		Periodic	1
		Frequent or with each suck	0
		Total Function score	

Management and treatment

The management of ankyloglossia can be either conservative or surgical. A “wait and see” approach is typically recommended for patients without significant functional impairment, as in some cases, it adapts over time.^{2,4} However, when ankyloglossia leads to feeding difficulties, speech issues, or other functional impairments, surgical intervention may be indicated. The choice of surgical technique depends on severity and condition, patient age, risk of recurrence, and prior treatment history.

Frenotomy is the simplest and least invasive procedure. It is often performed in neonates and infants experiencing breastfeeding difficulties. It involves a small incision along the midline of the frenulum, usually without suturing, making it a quick,

cost-effective outpatient procedure, though it carries a higher risk of recurrence due to contracture due to the absence of suturing.² (Figure 2)

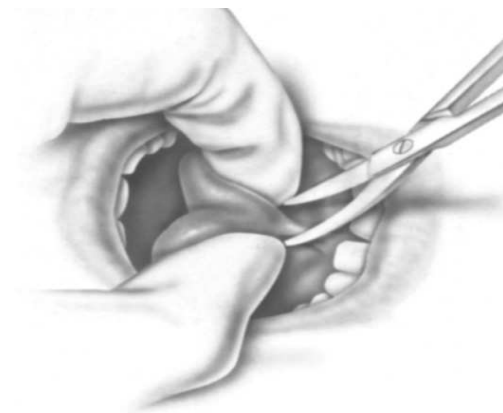


Figure 2. Frenotomy technique²

For moderate to severe cases with persistent functional impairment, frenectomy is preferred. This procedure involves complete excision of the lingual frenulum, with or without mucosal suturing, and can be performed under local or general anesthesia. The key principle of frenectomy is achieving a tension-free closure, which helps reduce contracture risk developing during the wound healing process. This procedure is more commonly performed in children by excising the entire lingual frenulum mucosa, and freeing the ventral tongue attachment through blunt dissection.^{2,9} (Figure 3)

However, some literature suggests that recurrence due to contracture can still occur, potentially caused by an imperfect tension-free

closure. Despite its complexity, frenectomy is a more comprehensive approach, aimed at minimizing recurrence, and improving outcomes in patients with ankyloglossia.^{2,9}

Frenuloplasty is a reconstructive technique used to address contractures or recurrences of ankyloglossia following frenotomy or frenectomy. The goal of frenuloplasty is to release restricted tissue and achieve tension-free mucosal closure using plastic reconstructive techniques that minimize scar tissue formation, and prevent re-contracture. Several frenuloplasty techniques can be performed to achieve better tension-free closure, including the transversal-vertical technique, Z-plasty with two flaps, and Z-plasty with four flaps.^{2,10}



Figure 3. Frenectomy technique⁹

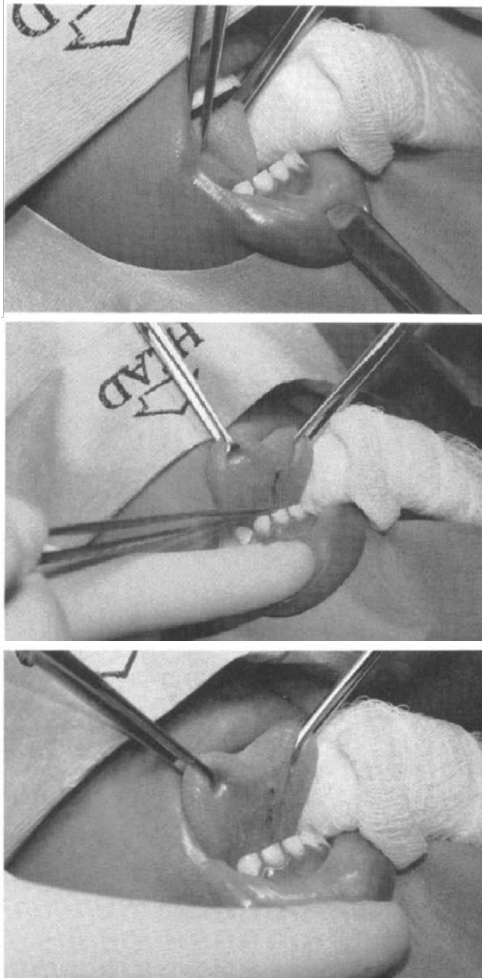


Figure 4. Transverse-vertical frenuloplasty technique¹⁰

The transverso-vertical frenuloplasty technique is particularly useful for cases with a restrictive lingual frenulum without significant scar contracture. It involves making a horizontal incision on the lingual frenulum at the base of the mouth, followed by blunt dissection to release the ventral tongue attachment, allowing adequate tongue mobilization. Once released, the mucosal edges are reapproximated vertically, and sutured with absorbable sutures. (Figure 4)

This method effectively converts the horizontal defect into a vertical closure, improving tongue mobility while reducing the risk of re-adhesion and scar contraction. However, it is less effective in cases with significant scar formation or recurrent contractures.^{2,10}

For more advanced cases, Z-plasty is the preferred technique as it improves tissue mobility, and reduces contracture risk. The two-flap Z-plasty involves a midline vertical incision along the frenulum, extending from the base of the tongue to the alveolar ridge, with two additional oblique incisions at angles of 45° to 60° at the ends of the vertical incision. These triangular flaps are then transposed and sutured using chromic 4-0 sutures. This flap transposition increases the length of the release tissue and redistributes tension, making it suitable for moderate cases where simple frenotomy or frenectomy has led to re-contracture. In severe cases, with significant fibrosis or multiple failed procedures, a four-flap Z-plasty (double Z-plasty) is preferred. This technique follows the same principles as the two-flap Z-plasty but includes two additional triangular flaps, providing even greater tissue elongation and mobility. The incisions are designed to redistribute tissue more effectively, reducing tension, and preventing recurrence. Double Z-plasty is particularly useful in severe ankyloglossia cases or recurrent cases with significant fibrosis, as it offers greater flexibility and functional restoration of tongue movement, especially in older children or adults.² (Figure 5, 6)

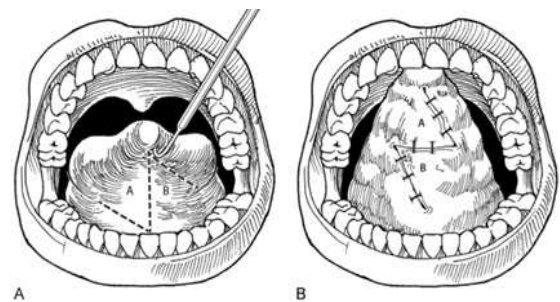


Figure 5. Frenuloplasty technique with Z-plasty²

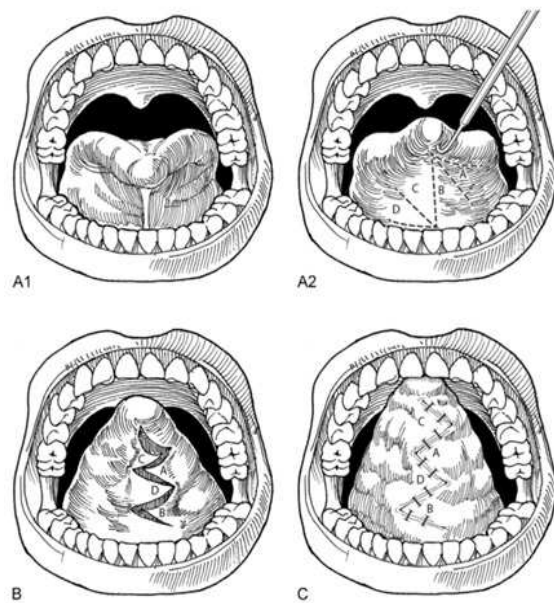


Figure 6. Frenuloplasty with double Z-plasty²

DISCUSSION

Ankyloglossia, or tongue-tie, is a congenital anomaly with wide-ranging clinical significance, from benign anatomical variants to condition that leads to impaired essential functions such as breast feeding, speech, and oral hygiene.^{2,3} While Kotlow's classification provides a useful anatomical framework, the degree of functional limitation remains variable and often subjective, complicating clinical decision making.⁴

Despite the literature favoring early diagnosis and intervention, particularly in infants with breastfeeding difficulties, controversy persist regarding the threshold for treatment.¹ While studies support the efficacy of simple frenotomy in improving latch and maternal comfort, outcomes are not universally positive.² Recurrence or inadequate symptom resolution highlights the need for careful case selection, structured follow-up, and parental counseling.

In older children and adult, the presentation shifts toward persistent articulation disorders, mandibular growth disturbance, and psychosocial distress. These cases often required more complex procedures

such as frenectomy or frenuloplasty, with techniques like Z-plasty showing potential in minimizing re-contraction, though data comparing their long-term outcomes remain scarce. Most available studies report follow-up periods of 6 to 12 months, limiting conclusions about sustained benefits or relapse rates beyond the first postoperative year.^{12,13}

One of the major challenges lies in the absence of a universal accepted diagnostic tools. Scoring system such as the Hazelbaker Assessment Tool (HATLLF) provides structured evaluations, but their predictive value for treatment success is inconsistent.¹¹ This diagnostic ambiguity may contribute to the wide variation in clinical practice, both in over-treatment of minor cases, and under-recognition of significant functional impairment.

Furthermore, cultural, parental, and practitioner biases may play a non-negligible role in treatment decisions. In many settings, pressure from caregivers to "fix" a perceived problem often based on social expectations, or feeding struggles. This can lead to early surgical intervention without clear indications.^{14,15} Conversely, in low-resource settings, access to skilled evaluation and surgical care is limited, resulting in untreated functional disabilities.¹⁶

Given these challenges, a structured, functional-based clinical algorithm is needed to aid decision making. Such an approach should integrate anatomical grading, functional assessment, patient age, and symptom severity. Additionally, there is a need to explore long-term developmental and psychosocial outcomes across different treatment modalities. Genetic and syndromic associations with ankyloglossia remain under-explored and could aid in early screening, especially in neonates with other congenital anomalies. Future research should prioritize standardizing diagnostic criteria, validating functional scoring tools, and assessing the

long-term efficacy of surgical interventions. Moreover, interdisciplinary collaboration among pediatricians, lactation consultants, speech therapists, and surgeons is essential to ensure holistic care tailored to each patient's developmental needs.

In conclusion, ankyloglossia is a common congenital condition with a wide range of clinical presentations. While many cases are mild and require no treatment, others can significantly affect feeding, speech, oral health, and social function. Early diagnosis and individualized treatment planning based on age, symptoms, and tongue mobility, are keys to effective management. Simple frenotomy is often sufficient in infants, but older or recurrent cases may need more complex surgical approaches. Standardized diagnostic tools and long-term outcome studies are still lacking, highlighting the need for further research and clearer clinical guidelines.

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