

ORIGINAL PAPER

## Ankyloglossia as an interdisciplinary problem

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### ABSTRACT

**Introduction:** The aim of the work is to present the current interdisciplinary knowledge on ankyloglossia and to initiate an interdisciplinary discussion in order to develop an algorithm of treatment.

**Material and methods:** The chairman of the Pediatric Otolaryngology Section at the Polish Society of ENT (ear, nose, and throat) appointed a working group of interdisciplinary experts of ankyloglossia in order to develop standards of treatment in children with a short lingual frenulum. This publication is a summary of speeches of participants of the meeting.

**Results:** The study presents the position on ankyloglossia in the fields of laryngology, neonatology, neurologopedia, dentistry, orthodontics and pediatrics. All authors unanimously emphasize the lack of sufficient, reliable research and clear guidelines on this topic. The basic issue regarding ankyloglossia is problems with maternal breast sucking, which often require frenotomy already in the neonatal ward. Doubtful cases are referred for more extensive diagnostics. Consultation with a neurologist along with myofunctional assessment of the tongue is crucial. Ankyloglossia can also make it difficult to expand the diet in small children, causing problems with swallowing and oral hygiene. It can also cause malocclusion and speech defects.

**Conclusions:** The interdisciplinary discussion enabled a broader look at the subject of the frenulum of the tongue and enabled the development of an algorithm for dealing with ankyloglossia, which will greatly facilitate the work of doctors, speech therapists and physiotherapists. The authors concluded that 3 groups of patients should be considered separately: neonates and infants, adolescents and adults. Many issues related to the frenulum of the tongue require further research, and knowledge on this subject will certainly evolve. However, it is crucial to develop rules of mutual communication and interdisciplinary cooperation, which may result in further progress in a more thorough understanding of this issue.

### KEY WORDS:

**ankyloglossia, frenotomy, guidelines in ankyloglossia.**

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## INTRODUCTION

Ankyloglossia currently raises a lot of controversy among specialists in fields such as pediatrics, laryngology, speech therapy, neonatology, physiotherapy, dentistry and orthodontics. Standards of treatment have been developed in a few countries, but they often differ from one another. In Poland, no unified recommendations regarding the management of this defect have been presented so far. This publication is an introduction to the development of interdisciplinary cooperation in ankyloglossia, with presentation of the current state of knowledge on ankyloglossia by specialists in this field.

## MATERIAL AND METHODS

The chairman of the Pediatric Otolaryngology Section at the Polish Society of Otorhinolaryngologists and Head and Neck Surgeons appointed a working group of experts from various fields in the subject of ankyloglossia in order to develop interdisciplinary standards in Poland. At the first meeting in May 2022, the participants presented their current, specialist knowledge on the subject, and the abstracts of the speeches are the content of this publication.

## RESULTS

The presented summaries reflect the current state of knowledge on ankyloglossia in particular fields. Therefore, they should not be regarded as guidelines. Standards of treatment in ankyloglossia will be developed after an interdisciplinary discussion at the next meeting of the working group.

### OPINION OF EAR, NOSE, AND THROAT SPECIALISTS

Ankyloglossia guidelines were developed by the American Academy of Otolaryngology – Head and Neck Surgery (AAO-HNSF) and published in 2020 [1].

1. The term ankyloglossia has been defined: a condition of the frenulum of the tongue that impairs tongue motility.
2. It was found necessary to conduct extensive, randomized studies on this subject and to establish interdisciplinary criteria for management.
3. Difficulties with suckling the breast and actions aimed at maintaining breastfeeding are the foremost problems. The diagnosis of ankyloglossia in the case of infants with difficulty suckling requires an “as soon as possible” frenotomy.
4. Contraindications to frenotomy are: Pierre-Robin syndrome and neuromuscular diseases.
5. A short frenulum of the tongue can cause an incorrect resting position of the tongue (at the bottom

of the mouth), which can result in jaw deformity and malocclusion.

6. Ankyloglossia can cause difficulties in articulation and in maintaining proper oral hygiene, which promotes tooth decay.
7. Previously published studies on the impact of ankyloglossia on speech disorders have poor credibility (small groups of patients, no randomized studies).
8. The decision to perform frenotomy in children with articulation disorders must be made after a thorough speech therapy assessment.
9. The frenotomy procedure is considered to be safe, with a low risk of serious complications.
10. Rare complications after the procedure include bleeding, airway obstruction, impairment to the ducts of the submandibular glands, eating reluctance and wound healing difficulties.
11. Before deciding on a frenotomy parents must be informed of the possibility of no improvement despite the procedure and of non-surgical alternatives.
12. The AAO-HNSF does not support precautionary frenotomy done to prevent possible sequelae of ankyloglossia, e.g. speech and malocclusion defects.
13. There is no age limit for this procedure, but it is recommended to avoid performing it under general anesthesia or without local anesthetics.

### COMMENT

1. The issue of anesthesia for the procedure remains contentious. For humanitarian reasons, care should be taken to ensure that no child suffers during frenotomy.
2. The decision on frenotomy and the type of anesthesia should be discussed with the child's parents, taking into consideration a number of aspects, not only regarding the child's health, but also the child's psychological well-being. Opinions of specialists justifying the need for surgical intervention should also be taken into account.
3. In children attending nurseries and kindergartens, the decision on frenotomy due to speech disorders should be preceded by hearing assessment due to the frequent coexistence of chronic exudative otitis, which is the most important cause of articulation disorders.
4. It is optimal to perform this procedure under general anesthesia, in conditions that are comfortable for the child and the operator, preferably comprehensively, e.g. during adenectomy or ear drainage. Therefore, the recommended specialists for the final qualification for frenotomy in children of nursery and pre-school age should be pediatric laryngologists.
5. Another dilemma is “preventive undercutting of the frenulum of the tongue”. Experts from the USA, in the quoted consensus, do not support such a procedure [1]. In turn, for example, specialists from Brazil

recommend frenotomy in all infants with a shortened frenulum of the tongue as a form of prophylaxis in relation to malocclusion and speech defects. Which of these options is correct? Undoubtedly for some patients one solution is good, for others the other. Only extensive, credible, clinical trials based on evidence-based medicine can solve this dilemma.

6. Post-operative care is an essential aspect of frenotomy due to the high tendency to form adhesions in the sublingual area. Thus, standards in this regard should be strictly defined.

#### OPINION OF A NEONATOLOGIST

In the neonatal period, in the case of 12–44% of children ankyloglossia causes difficulties in breastfeeding [2]. Therefore, this problem should be particularly taken into account by neonatologists in order not to give up on natural feeding. The standard used in the Department of Neonatology and Neonatal Intensive Care of the University of Silesia is presented below.

1. Identification of breastfeeding difficulties of the newborn and mother's difficulties with breastfeeding (painful feeding, sore nipples, milk stagnation, insufficient milk production) or on the part of the newborn (excessive loss or failure to gain weight, nipple biting, smacking noises during feeding, difficulty in grasping nipples, frequent interruption of sucking, reluctance to latch on) is an indication for a detailed assessment of the act of sucking using the Breast Sucking Skills Assessment Protocol [3]. To assess the frenulum of the tongue, the TABBY scale (Tongue Assessment Tool) is used, evaluating on a scale of 0–2: the appearance of the tongue, attachment of the frenulum to the gum, lifting the tongue with the mouth open and protruding the tongue [4]. The sum of the scores 0–3 indicates severe impairment of tongue function. Neonates with a score of  $\leq 5$  qualify for frenotomy.
2. Qualifications for frenotomy procedure: a restrictive frenulum (total score 0–3) should be cut as soon as possible after birth. In doubtful cases, the procedure can be postponed for a few days in order to correct possible mistakes in the technique of latching on, bearing in mind that the procedure not performed before the age of 4 weeks is associated with a high risk of interruption of breastfeeding. In the case of a frenulum more than 2 mm thick or the presence of abundant sublingual meatus, the procedure should be performed by an otolaryngologist or dentist.

Performing a frenotomy in the neonatal ward: Frenotomy is considered a safe procedure with a low risk of complications. The procedure requires the written consent of the parent. The incision should extend to the groove between the lower surface of the tongue and the floor of the mouth. It is recommended to massage

the sublingual area 4–6 times a day for several weeks after the procedure to avoid adhesions. After the newborn is discharged home, further consultation at the ear, nose, and throat (ENT) clinic is recommended. Contraindications to frenotomy: Pierre Robin sequence, decreased muscle tone, coagulation disorders.

Note: The finding of an abnormal anatomical structure of the tongue frenulum that does not lead to difficulties in breastfeeding is not an indication for frenotomy in the neonatal department. In such cases parents should be informed about the found anomaly and advised to examine the effectiveness of breastfeeding at home, in addition to a check-up with an ENT or neurologopedist, due to the increased risk of malocclusion and incorrect pronunciation [2].

#### STANDARD OF SPEECH THERAPY UP TO 3 YEARS OF AGE (SPEECH THERAPIST)

In infants with ankyloglossia problems may manifest not only when sucking on the breast and bottle but also when taking food from a spoon, biting, chewing or drinking from a cup. In some of the examined infants the following are observed: non-normative resting positions of the lips, jaw and tongue, snoring during sleep, episodes of apnea, poor ability to clean the tongue after feeding (white coating on the tongue), frequent regurgitation, choking on milk and saliva [5, 6]. Feeding problems in infants with ankyloglossia relate primarily to impaired sucking mechanics which may be manifested by poor cupping, retraction of the tongue, compensatory movements of the gums or placing the tongue in excessive protrusion. Infants with a short frenulum of the tongue may generate too low or too high a vacuum value during sucking [7]. The assessment of these patients cannot be based solely on visual assessment of the tongue and the location of the frenulum attachments. In the Silesian Infant Center children are examined on the basis of a protocol published in 2020, which focuses primarily on the assessment of oral functions [8]. English-language scales and protocols for the assessment of infants with a short frenulum of the tongue seem to focus on the suckling stage, ignoring the all-important stage of expanding the diet. Both the Hazelbaker scale and the later HATLFF scale modified by Amir do not refer to older children, in whom ankyloglossia is manifested by problems with expanding the diet [9, 10]. The Kotlow and Coryllos classifications are not functional assessment tools [11, 12]. The Martinelli protocol, despite being a tool for anatomical and functional assessment, does not provide guidelines for the functional assessment of an older infant [13]. An older infant with a short frenulum of the tongue does not always reach motor, sensory and reflex readiness on time to cope with foods of different consistency and structure at the stage of expanding the diet, which was included in the 2020 screening test [8].

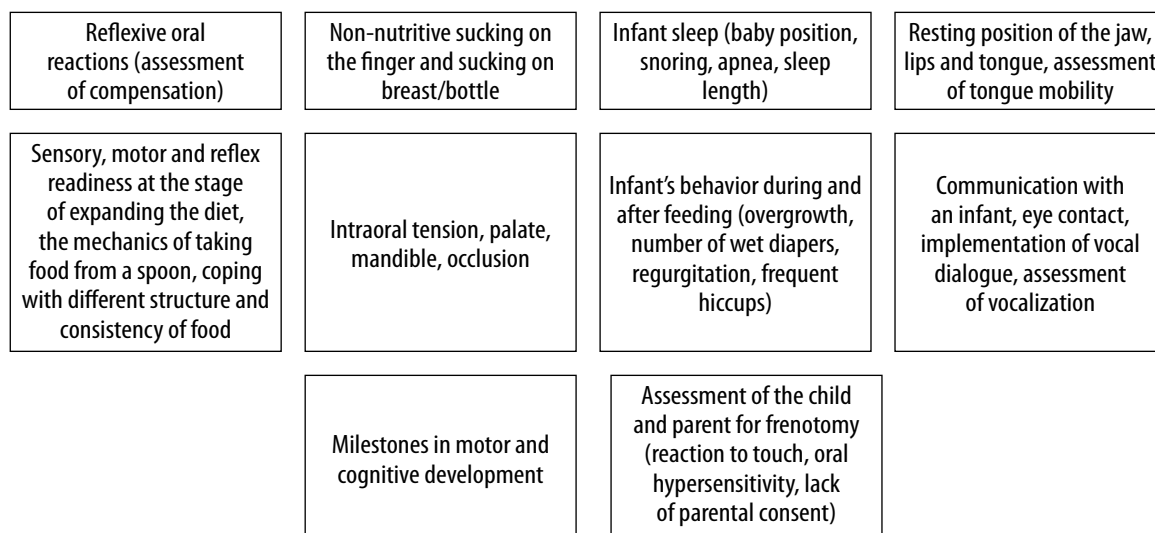


FIGURE 1. Diagnostic neurologopedic protocol for infants with ankyloglossia

## PROCEDURES

Based on the speech therapy protocol, the specialist evaluates, among other things, the resting positions of the oral apparatus, oral reflexes, sucking mechanics, sucking rhythm, swallowing-sucking ratio, and the infant's behavior during feeding. In older infants the ability to take food from a spoon, coping with the various consistency and structures of food, the mechanics of breathing and swallowing, the implementation of non-nutritive sucking on the finger, intraoral tension, and the assessment of compensatory movements are tested [8].

In the assessment of an infant with a short frenulum of the tongue it is necessary to take into account postural development and regulation of muscle tone. Intraoral assessment is insufficient, which is why many patients are referred to a physiotherapist for consultation. Due to abnormal tension patterns of the spine, skull base, soft tissues, as well as abdominal problems (colic, reflux), some infants are referred to an osteopath. Very often, before referring a child for a frenotomy consultation, we ask for the support of a certified lactation consultant. In infants with ankyloglossia we observe a number of adaptations and compensation mechanisms that allow for the implementation of the sucking function, despite the disturbed structure. The assessment, based on the speech therapy protocol published in 2020, includes the following areas that are monitored by a speech therapist [8] (Figure 1).

In a situation where a speech therapist/neurologist is convinced that rehabilitation alone will not sufficiently improve the function of the tongue with a shortened frenulum, he issues a written opinion and refers to a surgeon (ENT specialist, dentist, pediatric surgeon) with a recommendation to consider a frenotomy. In the Silesian Infant Center, we use the Parent and Therapist Notebook developed with Dr. Danuta Rup [14]. During the visit, the specialist shows the parent the exercises to be performed

before and after the frenotomy, which are individually tailored to the needs of the infant and the parent. With a written description of the diagnosis and recommendations, the child goes to the doctor-operator.

In the treatment of an infant with ankyloglossia, we include the following areas [8] (Figure 2).

The standard of the Silesian Infant Center is the monitoring of the postoperative period – parents send photos of the healing wound to a speech therapist and inform about the child's well-being. Selected exercises are modified for a specific child on an ongoing basis. About two weeks after the frenotomy there is a consultation with rediagnosis.

## DENTIST'S OPINION

The frenulum is an anatomical structure present in every human being, and only when it limits the mobility of the tongue should a surgical correction be considered. In infants aged up to 12 months, ankyloglossia can cause problems with feeding from both the breast and the bottle, as well as when the diet is being expanded. In the case of problems with breastfeeding it is necessary for the mother-child pair to be assessed by a Certified Lactation Consultant. The myofunctional assessment is performed by a neurologopedist. In case of disorders each child should have myofunctional therapy before and after the procedure of frenotomy. It aims to prepare the entire myofascial system of the orofacial area for proper mechanics and proper muscle tone. In the case of the anterior frenulum of the tongue (type I and II according to Coryllos) tongue mobility limitation is almost obvious. Patients with this type of frenulum experience functional limitations and muscular compensations. Posterior frenulum (types III and IV of Coryllos) cause greater diagnostic difficulties. It is because the symptoms that may result from the shortening of the frenulum coincide with

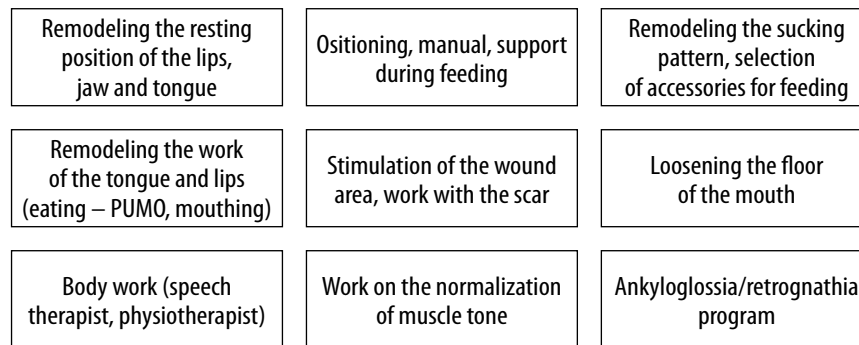


FIGURE 2. Areas of treatment for an infant with ankyloglossia

other dysfunctions of the child: disproportion in muscle tone, reflux, asymmetry, food allergies. Therefore, before the frenotomy the child should be holistically examined and the procedure itself should be considered only after excluding other disorders.

The procedure in my practice is: a neurologopedic assessment of the child, myofunctional therapy for a minimum of 2–3 weeks, rediagnosis of a neurologopedist with an assessment of the child's and parent's readiness for the procedure. For children with tension problems additional care of a physiotherapist or osteopath is recommended. After the frenotomy the parent is recommended stimulations in the oral cavity to restore the correct movement patterns of the tongue.

#### OPINION OF THE ORTHODONTIST

Since the “father of orthodontics” Edward Hartley Angle in 1907, there has been a constant debate on whether the incorrect position of the tongue is the cause [15–17] or the result of malocclusion [18]. Moss's functional matrix theory explained the mechanisms underlying the formation of malocclusions which laid the foundations for effective treatment in orthodontics. It assumes that the soft tissues of the orofacial area model the craniofacial skeleton in ontogenetic development [18]. The structure determines the function, but on the other hand a faulty function can change the development of anatomical structures [10, 18]. Orthodontists dealing with maxillofacial orthopedics very clearly notice the close connection between the lower, incorrect resting position of the tongue, often caused by a shortened frenulum of the tongue, and the development of such malocclusions as protrusion of the upper and/or lower incisors, partial anterior open bite and skeletal class III malocclusions [19–21]. In the case of ankyloglossia, the latest Polish academic textbook by Irena Karłowska entitled *Outline of Contemporary Orthodontics* recommends frenotomy [21]. The awareness of the importance of the correct structure of the tongue frenulum is usually limited to those orthodontists who are working closely with speech therapists. Unfortunately, the claim about the impact of a shortened frenulum on the malocclusion

is based mainly on subjective observation [20]. The lack of objective research on this issue results from the difficulties in developing a uniform, reliable classification of ankyloglossia and standards of management. It also makes it impossible to compare study results [19, 20]. The negative impact of a short frenulum on suckling in newborns is best documented. However, little research has been done on the relationship between ankyloglossia and malocclusion. These are single case reports and studies on small groups [16, 19, 21]. There are limited objective data confirming these relationships. Therefore, there is a need to create a Polish classification of the frenulum of the tongue to conduct further research based on it. It will help to explain the influence of ankyloglossia on the formation and development of malocclusion and to establish clear criteria for selection of patients for surgery.

#### ANKYLOGLOSSIA IN PEDIATRICS

The tongue is a very important organ. It affects suckling, swallowing, location, placement of teeth, periodontal tissue and speech [1]. Ankyloglossia is a genetically determined birth defect that is caused by insufficient apoptosis of cells in fetal life. This stops the process of separation of the tongue from the floor of the mouth [1]. The prevalence of ankyloglossia is 0.02–12% [22] with a male to female ratio of 2.5 : 1. This defect is distinguished using standardized tests in comparison with the results of laryngological examinations [23]. The review studies concerning the last 20 years indicate, in turn, the differences in the diagnosis of cases of ankyloglossia, as well as the methods of their treatment in different countries [24–28]. There are no rules of management in ankyloglossia in Poland. Therefore, there is an urgent need to create them.

#### ANKYLOGLOSSIA AND SUCKING PROBLEMS

Studies have shown that less than 50% of infants with ankyloglossia have problems with breastfeeding [29]. It should be noted that in recent years there has been a high variability and overdiagnosis of ankyloglossia in many countries, and too frequent, hasty referrals for frenotomy [23, 30–32]. Frenotomy is a procedure involving incision

of the shortened lingual frenulum to improve the mobility of the tongue. There are inconsistent data in recent review publications on the quality-improving effects of frenotomy. However, the conclusion was that frenotomy improves suckling by newborns and infants [31, 32].

#### ANKYLOGLOSSIA SPEECH THERAPY PROBLEMS

Messner *et al.* found that 60% of otolaryngologists, 50% of speech therapists and only 23% of pediatricians associate the occurrence of speech defects in children with the shortening of the lingual frenulum [33]. A serious problem in assessing this phenomenon is the fact that so far, no well-controlled, prospective studies examining the development of speech in a group of children with ankyloglossia have been conducted. This indicates that there is an urgent need to develop standards of ankyloglossia adapted to the organizational reality of Polish medicine. For pediatricians, it will be guidelines for proper management and supervision of the correctness of the care process for a child with a shortened frenulum of the tongue. The development of a clear algorithm of management will enable the fastest possible diagnosis and treatment of this defect and will eliminate the current confusion in this topic. It is therefore necessary to conduct prospective research with the participation of specialists dealing with this issue in the population of Polish children.

#### ANKYLOGLOSSIA AND MALOCCLUSION

It is known that the development of the maxilla and mandible, i.e. the facial skeleton, is dependent on the mobility of the tongue [34]. Dental problems such as deformation of the lower incisors, hypoplasia of the gums, and malocclusion in the context of shortening of the frenulum of the tongue as the cause have already been established and documented [35, 36]. However, there is insufficient evidence to promote prophylactic lingual ligament surgery to prevent malocclusion. It should be emphasized that this defect often coexists with other abnormalities in the upper respiratory tract, which should also be excluded [37, 38]. Therefore, before deciding on a frenotomy in a pediatric patient, comprehensive, interdisciplinary diagnostics should be performed first.

#### ANKYLOGLOSSIA AND ORAL HYGIENE

Dentists often diagnose ankyloglossia during their regular dental check-ups. One of the indications for frenotomy is improper oral and dental hygiene. Proper mobility of the tongue enables, among other things, the removal of pieces of food from between the teeth and ensures the proper movement of saliva in the mouth. This ensures proper cleaning and maintenance of the proper environment in the oral cavity (appropriate pH, bacteri-

al flora, enzymes, bactericidal substances). It also determines the initial grinding and digestion of food. The conducted research on this topic has confirmed the need to perform the procedure of frenotomy in patients with confirmed decreased oral hygiene as a result of limited tongue mobility [35]. In the absence of prospective studies, when preparing Polish recommendations/standards for frenotomy, one can only indicate the need to perform this procedure in such a group of patients, emphasizing, however, that these are not strong recommendations. Such an approach may seem justified due to the high incidence of caries in Polish children, where ankyloglossia may be one of the factors contributing to its development. Therefore, it would be worth proposing a health program under which each child with caries would have the condition of the tongue frenulum assessed in a dental office. It also seems reasonable to perform long-term prospective studies evaluating the impact of ankyloglossia and frenotomy on the condition of dentition in children in Poland.

#### CONCLUSIONS

The American Ankyloglossia Consensus is a summary and a valuable source of current, but incomplete knowledge about ankyloglossia. However, it does not propose a procedure algorithm that would facilitate interdisciplinary cooperation and systematize the practical knowledge necessary for doctors of various specialties. This justifies the need to create such standards and practical guidelines for professional groups interested in the subject. However, this requires an interdisciplinary discussion.

The difference in the point of view of different specialists, on the one hand, makes it difficult, but on the other hand, rationalizes the procedure and qualification for frenotomy.

At the meeting of the interdisciplinary group of experts the following conclusions were reached:

1. Standards of management in ankyloglossia should be developed independently for 3 groups of patients: A – newborns and infants, B – older children, C – adults.
2. The basic condition for interdisciplinary cooperation is the need to exchange information between specialists in particular fields in the form of written consultations.
3. It is recommended to perform photographic documentation of the sublingual area and assess the function of the tongue before and after the procedure.
4. The next meeting should be an attempt to agree and develop an interdisciplinary consensus in the form of an algorithm for dealing with a patient with suspected ankyloglossia in Poland.

#### DISCLOSURE

The authors declare no conflict of interest.

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