Article

Grouping of Ankyloglossia According to Coryllos Anatomical Classification and Follow-Up Results for Breastfeeding: Single Center, Cross-Sectional Study

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Abstract: Ankyloglossia is a condition of limited tongue mobility caused by a short lingual frenulum. The diagnosis and treatment of ankyloglossia are still controversial. The main clinical problem encountered during breastfeeding is difficulty in sucking and its clinical reflections. This study aims to evaluate the newborn population born regarding ankyloglossia and to determine the results of frenotomy. We conducted an observational, cross-sectional study among newborns born in a tertiary hospital. We included all newborns born between January 1 and June 30, 2022. The study algorithm was determined before the defined dates and data were recorded during the screening period. The recorded data were retrospectively collected from the files. Within six months, 705 babies were born. Due to additional problems and other conditions that prevent breastfeeding, evaluable data of 207 (29.3%) infants could not be provided. Of the remaining 498 infants, 234 (33.2%) had ankyloglossia. Breastfeeding was unsuccessful before frenotomy in 12 Coryllos type-1 patients, and all had difficulty in sucking. Frenotomy was performed within the three-months follow-up period in all patients with complaints of inability to firmly grasp the breast, nipple slipping from the mouth, and nipple biting during the first 24 hours. In terms of breastfeeding problems, regardless of the anatomical typology, frenotomy can be performed safely in the early life with successful results. If deficiencies or difficulties in nursing is noticed in ankyloglossia patients even at the first control, frenotomy should be recommended in clinical conditions.

Keywords: ankyloglossia; breastfeeding; Coryllos; frenotomy; lingual frenulum; newborn; tongue tie

1. Introduction

Ankyloglossia or tongue tie is defined as limited tongue mobility caused by a short lingual frenulum [1]. The lingual frenulum is a submucosal or membranous connective tissue component at the base of the tongue [2]. Abnormal development or incomplete dissociation of this ligament in congenital developmental stages results in ankyloglossia [3].

The diagnosis and treatment of ankyloglossia are still controversial [4-7]. According to the methods used for diagnosis, the incidence of ankyloglossia varies between less than 1% and 46.3% [8-10]. Sometimes, the diagnosis may get unnoticed, and patients encounter treatment delays and clinical problems; and in other instances, the patients may receive unnecessary interventions [11]. The main clinical problem encountered during breastfeeding is difficulty in sucking and its clinical reflections. Intervention is recommended as a consensus in patients with significant tongue movement limitation [1]. There are often no nursing problems even in this group of patients [1]. Even if breastfeeding is successful, problems may occur in tooth development, speech, and

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normal anatomical and functional development of the mouth and jaw structure at later ages [1,12].

This study aimed to characterize Kastamonu Training and Research Hospital newborn population according to the presence of ankyloglossia, determine its prevalence, characterize the types of ankyloglossia, decide whether or not it is clinically significant, and present the results of frenotomy procedures.

2. Materials and Methods

2.1 Study Plan

We conducted an observational and cross-sectional study of newborns born in a tertiary hospital that has approximately 1,500 deliveries per year and serves a population of roughly 300,000. We included all newborns born at Kastamonu Training and Research Hospital between January 1 and June 30, 2022. The study algorithm was determined before the defined dates and data were recorded during the screening period. The recorded data were retrospectively collected from the patient files.

2.2 Study Algorithm

Each newborn was subjected to neonatal examination by a pediatrician and pediatric surgeon. Corylloss groupings were noted in the neonatal examination records [13] (Table 1). Mothers of all babies born in Kastamonu Training and Research Hospital were given proper breastfeeding education and educational materials within national and international standards. Nutritional characteristics were recorded on the 1st, 7th, 28th, 60th, and 90th days postpartum by conducting face-to-face interviews with the mother, monitoring at least one feeding, and re-examination. Pain during breastfeeding, bruises and skin sores on the breast, and mastitis were evaluated and recorded. The infants' bad latch, inability to grasp the breast deeply, nipple slipping out of the mouth, nipple biting, suction power, suction amount, weight gain, and frequent and short-term partial nursing were also recorded [13].

Table 1. Types of Coryllos Ankyloglossia [13]

Type 1: Insertion of the frenulum to the tip of the tongue
Type 2: Insertion of the frenulum slightly (2 to 4 mm) behind the tip of the tongue
Type 3: Thickened frenulum attached to the mid-tongue and the middle of the floor of the mouth, usually tighter and less elastic
Type 4: Thick, shiny, and very inelastic submucosal frenulum that restricts movement at the base of the tongue

Other problems that may cause feeding problems in babies were re-evaluated. Mothers had received education in proper nutrition with breast milk one more time. Frenotomy was performed on babies with ankyloglossia who did not have any additional problems that would cause nutritional problems in clinical conditions. Babies who underwent frenotomy were followed up until they were 90 days old.

2.3 Frenotomy Procedure

All frenotomies were performed by pediatric surgeons in clinical conditions. There was no local or general anesthetic used. The babies were immobilized. The frenotomy was carried out by raising the tongue and cutting the frenulum up to the base of the tongue with blunt-tipped Metzenbaum scissors, allowing full exploration with a threaded sublingual ligament retractor. After the frenotomy, the patients were kept under observation for two hours. A health professional observed babies during their first feeding.

2.4 Statistical Analysis

Categorical variables (sex, method of delivery, prematurity, presence of ankyloglossia, clinically significant ankyloglossia, and Coryllos classification) were expressed as frequencies and percentages. We compared the populations with and without frenotomy. Qualitative variables were compared with the Pearson's chi-square test. Significance was set as $p < 0.05$. To perform statistical analyses, we used the IBM SPSS program version 22.

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3. Results

Within six months, 705 babies were born. Due to additional problems and other conditions that prevent breastfeeding, evaluable data of 207 (29.4%) infants could not be provided. Of the remaining 498 infants, 234 (33.2%) had ankyloglossia. Of the patients, 124 (52.0%) were boys, and 110 were girls (47.0%). Compared to 46 (19.65%) babies born by cesarean section, 188 (80.3%) were born by spontaneous vaginal delivery. Study groups were statistically similar in demographic characteristics, delivery method, birth weight, and sex (p>0.05).

Coryllos type-3 was the most common (70.5%) tongue-tie appearance. Frenotomy was performed in 67 patients due to clinical nursing difficulties caused by ankyloglossia. The need for frenotomy differed significantly between Coryllos groups (p<0.001) (Table 2).

Table 2. Coryllos groups and frenotomy distribution

	Frenotomy		Total	p
	Done	Not done		
Corylloss type 1	14 (6.1%)	0	14 (6.1%)	<0.001
Corylloss type 2	32 (13.7%)	12 (5.1%)	44 (18.8%)	
Corylloss type 3	15 (6.4%)	150 (64.1%)	165 (70.5%)	
Corylloss type 4	6 (2.5%)	5 (2.1%)	11 (4.6%)	
Total	67 (28.7%)	167 (71.3%)	234 (100%)	

Chi Square

The nutritional characteristics of the patients are given in Table 3. Breastfeeding was unsuccessful before frenotomy in 12 Coryllos type-1 patients, and all had difficulty in sucking.

Table 3. Nutritional Characteristics

		Type 1	**Type 2**	**Type 3**	**Type 4**	**Total**
Unable to Breastfeed	Day 1 (n=234)	12	8	3	0	**23**
	Day 7 (n=211)	0	0	0	0	**0**
	Day 28 (n=169)		0	0	0	**0**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Weak Grasp	Day 1 (n=234)	12	8	3	0	**23**
	Day 7 (n=211)	0	0	0	0	**0**
	Day 28 (n=169)		0	0	0	**0**
	Day 60 (n=167)		0	0	0	**0**

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	Day 90 (n=167)		0	0	0	**0**
Poor Latch	Day 1 (n=234)	14	32	15	6	**67**
	Day 7 (n=211)	2	24	12	6	**34**
	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Nipple Slipping Out of the Mouth	Day 1 (n=234)	14	32	15	6	**67**
	Day 7 (n=211)	2	24	12	6	**34**
	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Nipple Biting	Day 1 (n=234)	14	32	15	6	**67**
	Day 7 (n=211)	2	24	12	6	**34**
	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Weak Suction	Day 1 (n=234)	14	32	15	6	**67**
	Day 7 (n=211)	2	24	12	6	**34**
	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Weight Loss	Day 1 (n=234)	0	0	0	0	**0**
	Day 7 (n=211)	0	0	0	0	**0**
	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Frequent-Short Feeding	Day 1 (n=234)	14	32	15	6	**67**
	Day 7 (n=211)	2	24	12	6	**34**

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Durations	Day 28 (n=169)		0	0	2	**2**
	Day 60 (n=167)		0	0	0	**0**
	Day 90 (n=167)		0	0	0	**0**
Frenotomy Procedure	Day 1 (n=234)	12	8	3	0	**23**
	Day 7 (n=211)	2	24	12	4	**42**
	Day 28 (n=169)	0	0	0	2	**2**
	Day 60 (n=167)	0	0	0	0	**0**
	Day 90 (n=167)	0	0	0	0	**0**

The mothers of babies with ankyloglossia did not experience pain while breastfeeding, bruises and skin sores on the breasts, and mastitis.

In addition, frenotomy was performed in all patients who complained of inability to grasp the breast deeply in the first 24 hours, nipple slipping out of the mouth, and nipple biting during the 3-month follow-up period. On the other hand, babies who could latch since the beginning did not need frenotomy due to nursing problems in the following period, regardless of the type. There were no early or late surgical complications and no need for re-frenotomy in patients who underwent frenotomy. During the 90-day follow-up, the patients who underwent frenotomy did not experience feeding problems.

4. Discussion

The incidence of ankyloglossia in our study group was 33.2%. This rate is considerably higher than the 0.3% ankyloglossia rate in the study conducted by Çetinkaya et al. in 2011 examining general oral cavity anomalies [8]. In addition, different rates are seen in various studies, ranging from less than 1% to 46.3% [8,9,14,15]. These differences are due to varying diagnostic methods and criteria. For example, in the study of Maya-Enero et al., a similarly planned research to ours, the rate was 46.3% [9]. Maya-Enero et al. reported symptoms in 70.2% of patients [9]. In our study, the rate of symptomatic patients was 28.7%. The discrepancy in this result alone reveals that there is still no definite opinion. Especially in Corylloss Type 3 and Type 4 patient groups, the rates are higher than in other studies [8,14,15]. We determined the necessity of surgical intervention according to clinical features rather than the scales used [16-19]. Our results are consistent with the questions Manteli evaluated regarding function [18]. It is not certain that nursing problems will occur when ankyloglossia is present. However, frenotomy is beneficial if ankyloglossia is present in patients with feeding problems.

In study, we followed the patients for 90 days. This period is not enough to reveal all the consequences of ankyloglossia. However, the main finding in our study is that the complaints of babies who cannot latch, experience the nipple's slipping out of mouth, and nipple biting caused by ankyloglossia within the first 24 hours, continue regardless of type, and they need frenotomy for comfortable breastfeeding in follow-ups. Although our frenotomy results seem better compared to the literature, there is still not enough information about problems that may arise other than breastfeeding for a longer time [20-22].

Although some researchers asked mothers about maternal difficulties with breastfeeding, we did not observe such problems [1,23]. We can solve the problems of patients who bite nipples and cannot latch on effectively in the neonatal period using our bedside frenotomy procedure before breast damage ensues. Similarly, Messner's article of consensus in 2020 and in Emond's study in 2014 demonstrates that early frenotomy procedures prevent maternal problems [1, 24].

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In our ankyloglossia patients, breastfeeding problems disappeared after frenotomy. We had no complications in the frenotomy procedure, which has a low complication rate in the literature [20-22]. Frenotomy should not be avoided in infants with symptoms since it contributes to breastfeeding, and the complication rates are acceptable.

In clinical conditions, frenotomy can be performed more easily than other methods described [22]. As Coryllos mentioned in his publication in 2004, he used local anesthesia during the frenotomy technique [13]. We did not require the use of local anesthetics in our procedure. Similarly, in the study of Sethi et al., they did not use local or systemic anesthetics for infants [26]. Topical anesthetics may increase patient comfort in children [27]. If patients with ankyloglossia were not noticed during infancy until preschool or later, the procedure might need to be performed under general anesthesia [27].

The main limitation of our study is the short follow-up period since the patients may have some clinical complaints of ankyloglossia, especially after starting to speak. In addition, the effect of frenotomy could not be fully demonstrated since it was performed on all patients with clinical problems, and the nursing issues of the patients on who we performed frenotomy were fixed with follow-up appointments. Despite the extensive literature, there is no definitive diagnosis or treatment method for ankyloglossia. Therefore, like all other studies on ankyloglossia, our study has similarities with others and also a different approach to the subject. These types of differences limit the reproducibility of the study.

5. Conclusions

Concerning nursing problems, regardless of the anatomical typology, frenotomy can be performed safely in the early period to infants and can give successful results. If difficulties in nursing are noticed in ankyloglossia patients even at the first control, the frenotomy procedure should be recommended in clinical conditions. Randomized and controlled multicenter studies on large populations are necessary for diagnosing and treating ankyloglossia.

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Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: Data supporting the study are available from the corresponding author upon request.

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