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Treatment modalities in breastfeeding babies with marasmus and kwashiorkor associated with ankyloglossia



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ABSTRACT

Introduction: Successful breastfeeding depends on an infant's ability to latch onto the mother's breast correctly. A poor latch will cause insufficient milk transfer which will cause poor growth. Each type of mammal produces different components of milk which make it species-specific and adjusts to their needs, growth rate, and breastfeeding habits. Human breast milk has many benefits for human babies because it contains various substances both nutrition and protection. This study aims to present two infants who suffered from Ankyloglossia, which hindered the baby's ability to breastfeed and eat solid food.

Case Presentation: The study presented two infants who suffered from Ankyloglossia, which hindered the baby's ability to breastfeed and eat solid food. These babies were diagnosed with marasmus and kwashiorkor. After revision on both the lip- and tongue ties, supplementation with pasteurized human milk donor, and good complementary feeding, the babies finally reach optimal growth and development.

Conclusion: The sucking ability of the baby during early breastfeeding was proven to be important and should be examined, to achieve further successful breastfeeding.

Keywords: tongue-tie, lip-tie, kwashiorkor, marasmus, human milk donor, breastfeeding.

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INTRODUCTION

Human breast milk has many benefits for human babies, due to numerous beneficial substances. These substances, which are fat, protein, carbohydrates, mineral salts, and vitamins, serve as nutrition and as well as a protective factor for babies. Infants who receive sufficient breast milk are less prone to disease, due to the continuous exposure to the protective substances. These substances exist in the form of *Lactobacillus Bifidus*, lactoferrin, lysozymes, complement C3 and C4, antistreptococcal factors, antibodies, and cellular immunity, and do not cause allergies. Breastfeeding also provides beneficial psychological effects, leads to good growth, reduces the incidence of dental caries, and also reduces the incidence of malocclusion.¹

Each mammal has been provided with a pair or more breasts, which function to produce milk for their newborn. Milk produced by each mammal is species-specific, which is adjusted to its needs, growth rate, and breastfeeding habits.

For example, humans who are more resourceful and regarded as the most cognitively able mammal will certainly need breast milk that is of maximum benefit to the brain. Meanwhile, whales that live in a very cold environment and have a short feeding time, need milk with high-fat content so that they can stay warm in the cold oceans.¹

Breastfeeding is a natural thing, however, some difficulties may disturb a successful breastfeeding journey. Thus, a deep understanding of breastfeeding, both in terms of benefits and the technicalities of breastfeeding, is required. For a proper latch onto the breast, a baby needs to be well positioned on the mother's body and have a good mouth attachment to the breast. The position of breastfeeding can change according to the growth and development of the baby. Babies can be positioned to breastfeed lying on their back, lying on their side, sitting, or standing. The baby is held as close as possible and hugs the mother with both hands.¹

Good attachment to the breast in a

newborn is indicated by the baby's chin touching the breast, part of the areola inside the baby's mouth, the baby's lips folded out, and the mouth being wide open at least 130-160 degrees.² During sucking, the baby's mouth acts like a vacuum with negative pressure which sucks the breast, emptying its contents. This negative vacuum in the baby's mouth is affected by the lips that are folded outward, the tongue which can stick out past the lower gums, and the cheeks of the baby which are convex when feeding. If the vacuum is good, the suction is good, the baby will empty the breast properly, and the mother will feel that the breast is soft, not swollen, and there is no grip or irritation in the breast. A healthy baby can manage how much he needs to drink according to his body's needs.²

A lactation clinic located in Indonesia was used to obtain several cases of breastfeeding difficulties. The lactation doctor who works at the clinic is a member of the Praborini lactation team, a multi-disciplinary group of

professionals in Indonesia who have come together out of a common interest in the effective management of lactation. Founded in 2009, the team is composed of Pediatricians, Dentists, and Medical Doctors who are certified as Breastfeeding Counselors, IBCLCs (International Board Certified Lactation Consultants); led by Pediatrician, IBCLC. All doctors were trained by Perinasia (The Indonesian Society of Perinatology).³

During daily observation in the lactation clinic, some difficulties exist which can interfere with the latching of the baby's mouth and sucking on the breast. The position of the baby during pregnancy in the mother's stomach or the difficult and traumatic process of labor can inhibit the baby's suction reflex. Anatomical abnormalities in the baby's mouth can also interfere with suction, such as cleft lip or palate, ankyloglossia/tongue-tie, or lip tie. To suck properly, anatomical abnormalities in the baby's mouth need to be corrected. Among them employing frenotomy (cutting/incision) on the tongue or lip tie.²

This cutting method is known as Frenotomy using the Pare Method. In the 17th century in Italy, almost all babies who were baptized would have a frenotomy because the procedure would make it easier for the baby to latch onto the breast. The Pare method is used to cut the middle part of the tongue frenulum and free the bonds with the fingers to the genioglossus muscle. This action is safe and only takes a few seconds. Immediately after the frenotomy, the baby's skin turns pinker, the voice becomes clearer, the body temperature increases, and the baby becomes more relaxed, thus making breastfeeding easier.⁴

Malnutrition is estimated to cause death in one-third of child deaths in the world. Lack of access to nutritious food, wrong feeding practices such as inadequate breastfeeding, and inadequate complementary foods, could affect the nutrition profile of a child, which leads to malnutrition. Infectious diseases such as persistent diarrhea, pneumonia, measles, or malaria can affect the nutritional status of children.⁵

In children, Energy Protein Malnutrition is defined if the measurement

is below a -2 Standard Deviation. If body weight according to age is below -2 SD the child could be defined as underweight. Furthermore when the height is below -2 SD, can be defined as stunting and if weight according to height is below -2 SD, can be defined as wasting. Wasting indicates recent weight loss, while stunting is usually the result of chronic weight loss. Severe malnutrition can take many forms, for example, kwashiorkor and marasmus. Marasmus was caused by severe wasting conditions. Subcutaneous fat and muscle mass are lost due to a lack of nutrition. Marasmic kwashiorkor is a severe wasting condition with edema (accumulation of fluid between the body cell spaces), and kwashiorkor, which is malnutrition with edema, usually accompanied by changes in hair and skin color, anemia, hepatomegaly, lethargy, decreased immunity, and even death.⁶

According to the previous publication "A Holistic Supplementation Regimen for Tongue-Tied Babies with Slow Weight Gain and Failure to Thrive", it was found that Tongue- and lip-tie could prevent successful latching and prevent adequate nutrition. Tongue- and lip-tied babies can either have slow weight gain (SWG) or failure to thrive (FTT) and also low milk supply mothers. During the previous cross-sectional study of 55 tongue- and lip-tied babies with SWG and FTT, it was shown that the holistic management of tongue- and lip-tied babies with SWG or FTT consisting of frenotomy, at-the-breast supplementation, domperidone, and acupuncture could improved infant nutritional status and the mother's milk supply. Babies could breastfeed without supplementation after treatment and gain weight. In the previous study, 11 subjects with FTT did not gain adequate weight. These infants with FTT had excessive weight loss during early life due to poor milk transfer from tongue- and lip-tie and frenotomy and supplementation with feeding tube devices failed to adequately compensate. The lactation team provided early complementary feeding to these subjects. This approach was successful, and supplementation with feeding tube devices could later be stopped after improvements with breastfeeding and complementary feeding.³

Here we presented two infants who suffered from Ankyloglossia, which hindered the baby's ability to breastfeed and eat solid food. These babies were diagnosed with marasmus and kwashiorkor. After revision on both the lip- and tongue ties, supplementation with pasteurized human milk donor, and good complementary feeding, the babies finally reach optimal growth and development.

CASE PRESENTATION

Case 1: Breastfeeding and Marasmus

The baby in case 1 (Baby girl Hn) was born at 39 weeks of gestation via normal delivery. Early initiation of breastfeeding was not introduced, and rooming-in was started 13 hours after delivery. Soon she had latching and feeding difficulty. At one month of age, she was referred by the Indonesian Breastfeeding Mothers' Association to come to the lactation clinic due to the failure to thrive diagnosis.

Baby girl Hn was born weighing 3200 grams, however, at the age of one month, the baby weighed 1860 grams despite continued breastfeeding. Baby girl Hn looked very skinny, showing signs of subcutaneous fat and muscle mass loss. She was on < -3 standard deviation of the WHO growth chart. During the physical examination, both submucosal tongue-tie and upper lip-tie grade 3 were found. A breast examination of the mother revealed a symmetrical breast shape, prominent nipples, and a low milk supply. During breastfeeding, the baby's lips were folded inward and only the tip of the nipple went inside the baby's mouth, thus the suckling was slipping and the suction reflex was weak.

The mother (Mrs. U) agreed for her baby to undergo a frenotomy process and further supplementation with formula milk while waiting for a human milk donor through a supplementary nursing system. The mother was prescribed with breastmilk supplement which contains Galatonol, a bioactive fraction from *Sauropus androgynus* and *Coleus amboinicus*, in combination with *Striatin*, a bioactive protein fraction from *Channa striata*. The study demonstrates that the combination of Galatonol with *Striatin* has galactagogue properties because it can stimulate milk production, prolactin, and

Table 1. Baby Girl Hn's Development Upon Review to Pediatrician, IBCLC, Specialist of ADEL

Date	Age	Weight (gram)	Weight Gain (gr/day)	Nutritional Status	Human Milk Donor in SNS (cc/day)	Therapy
26/5/2020	1 months 13 days	2105	23,5	< -3SD	200-300	- Minimum pasteurized donor's breast milk in SNS: 300 cc/day - Home visits oral motor exercise every 2 days
	1 m 23 d	2365	26	< -3SD	280-300	- BCG vaccination - Min. pasteurized donor's breast milk: 360 cc/day - Mrs. U given extra maternity leave letter for 2 months
16/6/2020	2 m 3 d	2740	34,1	< -3SD	165-395	- Min. pasteurized donor's breast milk: 400 cc/day
30/6/2020	2 m 18 d	3250	36,4	< -3SD	375-435	- DPT – Polio – Hib – Hep. B vaccination - Min. pasteurized donor's breast milk: 400 cc/day - Home visits oral motor exercise stop
14/7/2020	3 m 1 d	3760	36,4	< -3SD	350-450	- Min. pasteurized donor's breast milk: 400 cc/day
30/7/2020	3 m 17 d	4130	23,1	< -3SD	250-370	- DPT – Polio – Hib – Hep. B vaccination - Min. pasteurized donor's breast milk: 350 cc/day
13/8/2020	4 m	4665	38,2	< -2 SD	245-370	- Min. pasteurized donor's breast milk: 350 cc/day
27/8/2020	4 m 13 d	4910	17,5	< -2 SD	220-385	- Baby H given early complementary feeding. - Breastfeeding on Baby H's demand using SNS contain pasteurized donor's breast milk
10/9/2020	4 m 26 d	5160	10	< -2 SD	60-135	- Breastfeeding on Baby H's demand using SNS contain pasteurized donor's breast milk - Increase frequency of complementary feeding
24/9/2020	5 m 11 d	5655	35,4	Good	55-150	- DPT – Polio – Hib – Hep. B vaccination - Breastfeeding on Baby H's demand using SNS contain pasteurized donor's breast milk - 1 month extra maternity leave letter for Mrs. U
8/10/2020	5 m 25 d	6050	28,2	Good	50-100	- Stop using SNS. - Complementary feeding and direct breastfeeding on demand or cup feeding when Mrs. U at work - Baby H given D vitamin and Fe

**Figure 1.** Baby Girl Hn's Development Serial Photographs

oxytocin release in female rats.

The mother was also taught how to do a routine lip and tongue exercise after the frenotomy. Three days after the procedure, both mother and baby came for a review.

There was no significant improvement with Baby Girl Hn, she was still sleepy all the time and did not do much drinking. During the observation she left much of the formula inside the SNS, thus the

body weight only increased from 10 grams to 1870 grams for 3 days. Further suggestion Mrs. U and Baby girl Hn was to be hospitalized to optimize the treatment.

During hospitalization, the mother and baby were advised to do skin-to-skin contact for 24 hours. The baby was given Piracetam 3 x 20mg and further breastfeeding was done with SNS filled with formula milk minimum of 6 x 30cc. Baby girl Hn was immediately consulted to the physical medicine and rehabilitation unit to be given Oral Motor Exercise therapy. After seven days of hospitalization, the body weight increased to 2065 grams, and allowed to go home. Further instructions to Mrs. U were to continue breastfeeding on demand assisted by SNS filled with pasteurized human milk donors and the consumption of breast milk supplement tablets was continued. For Baby Girl Hn, piracetam treatment was continued and the oral motor therapy was continued with daily home visits.

For several months, Baby Girl Hn continued to breastfeed with SNS filled with pasteurized human milk donors. Her laboratory results were within normal limits, except for the hemoglobin, which was low. Early complementary feeding was

started when baby girl Hn was at 4 months and 13 days of age. At 5 months and 11 days of age, she has reached good nutritional status. Baby girl Hn is still breastfeeding until this article is written. The progress of Baby Girl Hn during treatment can be seen in Table 1, Figure 1, and 2.

Case 2: Bottle Feeding, Goats Milk, and Kwashiorkor

The baby (Baby girl Hf) was born at 39 weeks of gestation via normal delivery. Early initiation of breastfeeding was introduced and the family was discharged from the hospital with adequate breastfeeding knowledge. However, at home, Baby girl Hf always cried and was

not satisfied during breastfeeding. From the age of 1-month-old, the mother (Mrs. Y) was advised by her friend to feed the baby with powdered goat's milk by bottle, apart from breastfeeding, and to avoid formula milk due to its sweetness. Baby girl Hf then had a breast rejection and preferred bottle-feeding. After the introduction of goat's milk, Baby girl Hf had multiple rashes on her skin and was hospitalized several times due to allergic reactions. Moreover, baby girl Hf also did not receive any vaccination.

When the complementary feeding was started at the age of 6 months, Baby girl Hf had feeding difficulty, in which she always vomited while eating and hard to swallow mashed food. As advised by a friend, the mother brought baby girl Hf to the lactation clinic to find out the root cause of the problem.

Baby girl Hf looked very sick, with edema all over her body, including her face, stomach, genital, and all extremities. When presented at the age of 9 months 1 week, she was not able to roll, creep, crawl, or sit. Her extremities felt cold. She could only moan with limited limb movement.

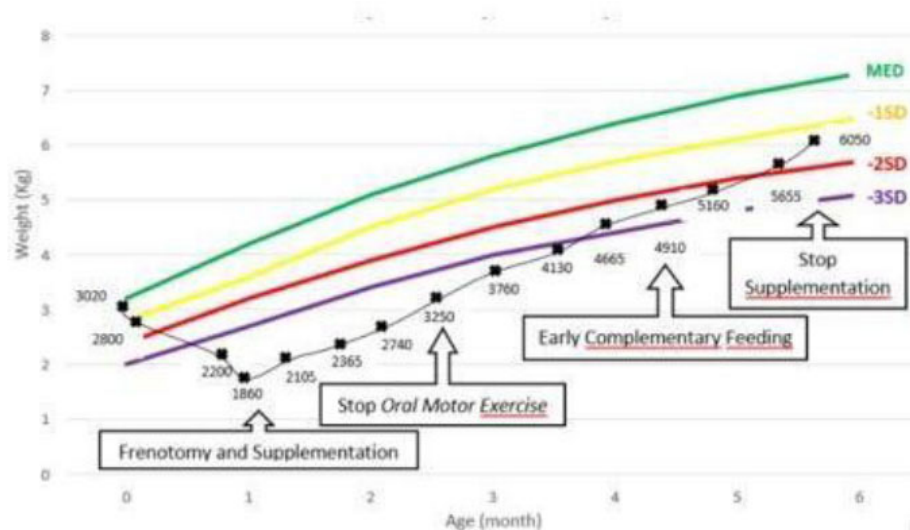


Figure 2. Baby Girl Hn's Development Chart.

Table 2. Baby Girl Hf's Development Upon Review to Pediatrician, IBCLC, Specialist of ADEL

Date	Age	Weight (gram)	Weight Gain (gr/day)	Nutritional Status	Development	Therapy
11/4/2020	9 months 7 days	4720	6,3	< -3SD	Global Development Delay	Lingual Frenotomy Tongue exercise 5 times per day by mother
18/4/2020	9 m 14 d	5585	123	< -3SD	Extremities warmer, some edema gone	Upper lip tie frenotomy Tongue and lip exercise 5 times per day by mother
25/4/2020	9 m 21 d	4610	-139	< -3SD	drink and eat more, the baby's swelling shrinks and the skin peels off	Vaccination of DPT, Hib, Polio, Hepatitis B, INH prophylaxis and iron supplementation. Tongue and lip exercise stopped
30/5/2020	11 m	6205	47	< -2SD	Sit and roll	MR vaccine Prophylactic INH, and iron supplementation
20/6/2020	12 m	6895	37	< -2SD	Crawl	Vaccination of DPT, Hib, Polio, Hepatitis B, Prophylactic INH, iron and vitamin D supplementation.
25/7/2020	13 m	7650	23,1	Good	Babbling	Vaccination of DPT, Hib, Polio, Hepatitis B INH prophylactic, iron and vitamin D supplementation.



Figure 3. Baby Girl Hf's Development Serial Photographs.

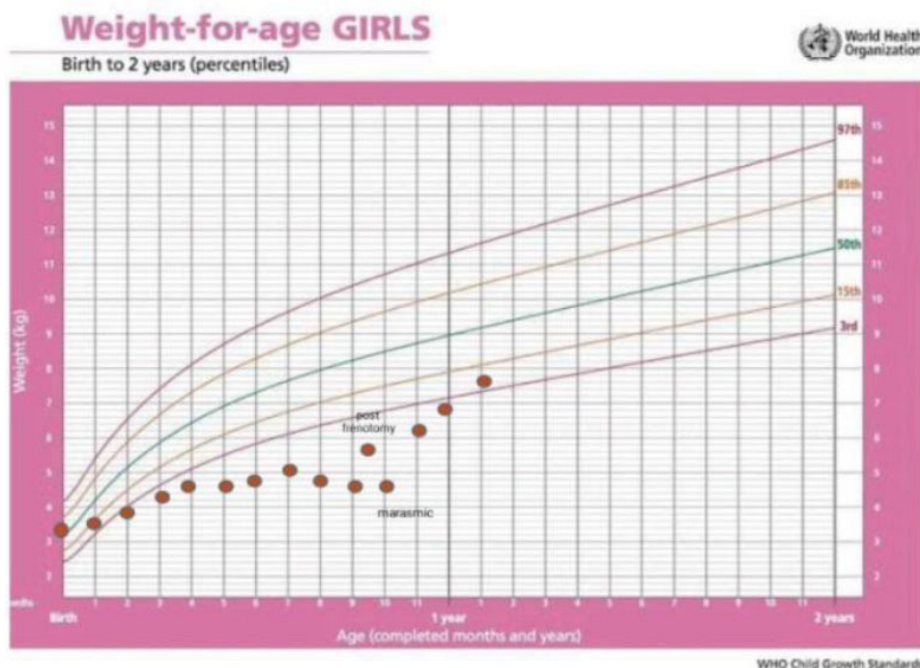


Figure 4. Baby Girl Hf's Development Chart.

Baby girl Hf was born weighing 3000 grams. At the age of 9 months and 1 week, she weighed 4720 grams despite continuous bottle-feeding and consuming a small amount of food. She was on < -3 standard deviation of the WHO growth chart. Submucosal tongue-tie and upper lip-tie grade 3 was found during physical examination. During the breast

examination, the mother had a protruding nipple with no milk supply. Mother was pregnant with a gestational age of 5 months.

The parents were counseled on the benefits of breast milk and the importance of human breast milk compared to goat's milk. The parents were also advised to look for human milk donors as

supplementation was needed to catch on with the weight loss. In the end, the parents were informed about tongue-tie, lip-tie, and frenotomy. The parents agreed to the correctional treatment and the frenotomy was performed.

Soon after the frenotomy was done, the baby was able to eat a corn cream soup without gagging and vomiting. The baby could also drink directly from a glass without gagging. Furthermore, the baby was encouraged to drink pasteurized human milk donor using a bottled teat and to stop goat's milk consumption as soon as the mother found a breast milk donor. A breastfeeding re-lactation was not encouraged, due to the mother's current pregnancy status and the baby's suckle difficulty. The mother was taught how to do tongue exercises 5 times a day for 3 weeks and tummy time.

For several months, baby girl Hf continued to drink pasteurized human milk donors through bottle teat and complementary feeding. At 13 months of age, she had reached a good nutritional status and thus, a good development. Until now, Baby girl Hf is still in good development status and is already able to walk at 15 months of age. The diagnosis was purely determined by the physical examination and clinical experience of the doctor who handled this case. The progress of Baby Girl Hf during treatment can be seen in Table 2, Figure 3, and Figure 4.

DISCUSSION

Successful breastfeeding depends on an infant's ability to latch onto the mother's breast correctly. A poor latch will cause insufficient milk transfer which will cause poor growth. In both cases, all babies experienced difficulty latching. Health personnel did not detect her tongue tie and lip tie in the first place. Thus, baby Hn was referred by the Indonesian Breastfeeding Mothers' Association to come to the lactation clinic due to the failure to thrive diagnosis, while baby Hf came to the lactation clinic with severe malnutrition after difficulty of breastfeeding and eating solids.¹

The difficulty of these cases is how to sustain both babies to breastfeed for up to two years whilst they were in severe

malnutrition. The Praborini team will do counseling not just from the scientific area but also from the patient's religion. In Indonesia, religion is part of the citizen's daily life. The Indonesian government recognizes six religions: Islam, Protestantism, Catholicism, Hinduism, Buddhism, and Confucianism. And all holy books of these religions include breastfeeding in their sentences.³

In case one, baby Hn's parents were so thrilled with the treatment and they were committed to continue breastfeeding for up to two years, first with a supplemental nursing system filled with human milk donors then early complementary feeding. In case two, baby Hf was receiving bottles from the age of 1-month-old, so that when she came at 9 months of age she was already in a state of nipple confusion, her mother was pregnant at that time and there was no milk supply. So the option for her is to receive a human milk donor from a bottle. Human milk produces different components of other species of milk (she received goat milk before coming to the lactation clinic).^{4,7}

With human milk donors, all babies' conditions significantly improved. Not just the baby's growth but also development. Baby Hf was only able to lie down for 9 months, but after receiving a human milk donor she can catch up with her growth and can walk at 15 months of age. Unfortunately, in both cases, health personnel at the health facility where the babies were born could not detect breastfeeding abnormalities in the first place. The baby who experiences breastfeeding difficulty may benefit from clinical oral examination and breastfeeding examination to seek tongue-tie and lip-tie.²

In these cases, after revision of both the lip- and tongue ties, supplementation with pasteurized human milk donor, and good complementary feeding, the babies finally reach optimal growth and development. The challenges of these cases are that we couldn't order detailed laboratory tests to mark patients' malnutrition because of the financial condition of parents. All consultation fees for these patients were waived by the treating pediatrician.

CONCLUSION

Human breast milk contains many beneficial ingredients to provide infants with sufficient nutrition for their growth and development. Difficulties may arise to disturb a successful breastfeeding journey, such as the baby's ability to latch on correctly to the mother's breast. The ability to detect breastfeeding problems needs to be known both by mothers and medical personnel. During the study, it was observed that by revising the ties, a reduction or elimination of breastfeeding problems could be achieved, thus preventing babies from disease or malnutrition. In the end, the sucking ability of the baby during early breastfeeding was proven to be important and should be examined, to achieve further successful breastfeeding.

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ETHICAL APPROVAL

The publication has been approved by the parents of the babies in the manuscript

CONFLICTS OF INTEREST

There is no conflict of interest.

AUTHOR CONTRIBUTION

All authors contributed equally to this study.

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