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Changes in Breastfeeding and of Breasts before and after Babies' Surgeries for Ankyloglossia with Deviation of the Epiglottis and Larynx (ADEL)

—Healthy Breastfeeding

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Abstract

Background: Historically frenotomy was performed at newborn babies' baptism for healthy breastfeeding over many centuries. Although it was necessary for nursing babies, its application was denied subjectively without clinical observation since the early 20th century. As a result solutions for breastfeeding and breast problems are confused now. Methods: We studied changes in breastfeeding and mothers' breasts before and one month after the surgeries for ADEL by standards for healthy breastfeeding and breasts. Results: With regard to suckling, before the surgeries 24% of babies opened their eyes during breastfeeding, while after surgeries 76% opened eyes while nursing. 30% of the babies latched onto the mother's breast with a wide-open mouth. After surgeries, 84% properly latched on. 73% of the babies had calluses on the upper lip before surgeries, and no calluses were observed after surgeries. 27% of babies before surgeries moved the jaws during breastfeeding, but after surgeries 88% moved the jaws while nursing. White debris was on the dorsum tongue in 70% of babies prior to surgeries. After surgeries, 22% were with debris. 67% of mothers experienced nipple pain before their babies' surgeries while 6% had pain after the surgeries. Flattened nipples were observed in 55% of mothers before the infants' surgeries, 7% after surgeries. 23% of mothers had breast pain before the surgeries, and 4% had breast pain after the surgeries. Mastitis was also reduced from 9% to 1% after surgeries. 73% of women had palpable breast masses, and after the surgeries 25% had palpable masses. Summary: Remarkable improvements in breastfeeding and in women's breasts were achieved after their infants underwent operations for ADEL. These results indicate the necessity of surgical interventions in babies with ADEL for healthy breastfeeding and breasts (279).

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Keywords

Correction of the Glosso-Larynx (CGL), Expansion of the Vestibular Oris (EVO), Nipple, Eye Contact, Respiration, Sleep, Snoring, Crying, Occlusion

1. Introduction

When I (S. Mukai) started as a physician specializing in otorhinolaryngology (1970), frenotomy was performed for a baby with ankyloglossia in Japan. The method used was that of Paré's frenotomy [1]. In the 17th century, in Italy almost all babies underwent frenotomy at their baptism whether or not a frenulum was present (Hieronymus, 1537-1619) [2]-[4]. The procedure was necessary for breastfeeding (Baudelocque, 1745-1810) [5].

Technique of Paré is to cut the center of the frenulum and separate the tongue with the finger pressed to the genioglossus muscle. It takes only a few seconds with little loss of blood and is safe. Immediately after the frenotomy the baby's complexion changes to pink, its voice becomes clearer, the body temperature rises, and the extremities become suppler. A baby with a frown on its face assumes a sunny expression and eye contact becomes possible. Breastfeeding as well as bottle feeding and childcare become easier after Paré's method.

These changes suggest that the tongue has a close relationship with the respiratory system. I observed the larynxes of babies with ankyloglossia by laryngeal fiberscope. The observations revealed that these babies had an up and forward deviation of the epiglottis and larynx. Pulse oximetry revealed that their SpO_2 was low and heart rate was high during breastfeeding. In addition they presented with a multitude of symptoms and signs such as hard crying, cold extremities, swollen abdomen, piloerection and/or alopecia, shallow sleep, snoring, etc.

Ankyloglossia with deviation of the epiglottis and larynx (ADEL) was named for these conditions instead mere ankyloglossia [6]-[11]. Correction of the glosso-larynx (CGL) and expansion of the vestibular oris (EVO) were employed to cure ADEL [12]-[15].

At a physical examination of 4-month-old babies at the Hodogaya and Izumi ward medical centers in Yokohama, 93.6% of the babies presented with ankyloglossia [16]. It was rather natural that almost all babies received frenotomy by midwives through the 17th till the early 20th centuries in Europe. Although modern medicine denied the need for frenotomy, human physiology had not changed [3] [17]. Humans still had ankyloglossia at a high rate [16].

Besides numerous symptoms and signs of ADEL, babies with ADEL have suckling problems. While breastfeeding, these infants hurt their mothers' breasts.

In 1993, Suzuki H. *et al.* submitted a paper "Relationship between babies with ankyloglossia and disturbance of their mothers' breast" to Journal of Human Lactation (JHL). A comment from JHL was as follows; (*Commenter C*) *I must reject this paper for publication. The definition of ankyloglossia is never clearly stated*; *if it is* "*ankyloglossia with deviation of the epiglottis and larynx*", *in my* 30 *years of experience as a practicing pediatrician and breastfeeding consultant, I must say that I have never seen a case. I wonder therefore, even considering the possible cultural differences, how the authors could have amassed* 162 *babies in one maternity clinic in a two-year period. The technique of frenotomy, after Pare, with* "*subsequent separation of the tongue as far as the genioglossal muscles, etc*." *would be very invasive and disfiguring and radical, and to my mind totally inappropriate to this simple clinical situation; simple bloodless, non-invasive frenotomy is curative. And—page* 6, *—normal suckling movement do NOT involve biting and chewing*; *if* "*babies with ankyloglossia… are reduced to sucking* (*sic*) *and swallowing*", *then they are doing right thing. Also, the authors seem to miss the point when they quote as abnormal that* (*p*7, *top*) *AB babies* "*suck the nipple by pressing it with the dorsum of tongue against the hard palate… and as a result their mothers*" *nipples change shape. Of course, ALL NORMAL babies suckle this way. This paper has many inaccuracies, and thus loses validity* [18].

I think the reasons for rejection were too subjective. As denials of Paré's frenotomy about hundred years ago there were several reports (2, 3, 4).

I read an article recently by L. Dahl in "Clinician's guide to breastfeeding (2015)". In her preface it was written; *When breastfeeding doesn't happen easily, it can be due to a simple problem, such as an improper latch or sleepy baby. But often only produce a few ounces of milk… the baby takes leisurely hour-long feeds, then gets hungry an hour later… the baby cries at the breast and becomes frantic… the mother experiences pain every*

time her baby latches on. Talk to a mother with cracked, bleeding nipples, and watch her cringe at the mere thought of her crying baby's mouth, and you'll see how bad it can get.

I discovered these symptoms myself with the birth of my daughter 12 *years ago, while I was at my residency. Memories of that time are blur of exhaustion and frustration. Lucy nursed almost constantly, every hour for* 45 *min. My nipples were raw, cracked and very painful, but because my baby seemed healthy, I thought this was normal. Lucy's pediatrician and my own doctor both told me everything was fine and I should just "wait out" the pain—it would stop eventually. She was gaining weight slowly, but within normal parameters.*

Over time, the art of breastfeeding was lost. It seems reasonable nowadays to ask your doctor for advice, but what doctor knows anything about the breastfeeding (pp vii-xi) [19].

Why has medicine lost proper views on healthy breastfeeding of babies and healthy breasts? Several reasons can be considered. 1) Majority of babies have ankyloglossia, as seen by the examination of 4-month-old babies described above. 2) Relationship between ankyloglossia and respiration is overlooked. 3) Symptoms and signs of ADEL are also overlooked. 4) The need for Paré's frenotomy is denied without correct clinical observation but subjectively. 5) I think that the most important point is that there is no standard for healthy breastfeeding.

Babies with ADEL hurt their mother's nipples and breasts by breastfeeding. We have already reported changes in breastfeeding and the breasts before and one week after CGL and EVO [20]. For the current study changes in breastfeeding and mother's breasts before and one month after CGL and EVO are described as are healthy breastfeeding and breasts.

Studied items are as follows:

1) Changes in breastfeeding by the baby.
2) Changes in mother's nipples.
3) Changes in mother's breasts.

2. Cases

From Dec. 3, 2014 to Nov. 28, 2015, 67 infants who were diagnosed as having ADEL and their mothers were studied. All babies were breastfed. Age distribution of the babies was from 11 days to 4 months and 20 days (average age: 2 months and 1 day). Age distribution of their mothers was not studied. All of these babies had undergone CGL and EVO.

3. Methods

3.1. Studied Items

The following items were studied before and one month after the babies' CGL and EVO: (A) suckling by the babies, (B) changes in mothers' nipples, and (C) changes in mothers' breasts.

(A) Suckling by babies

In healthy breastfeeding, the baby opens its eyes wide and latches onto the mother's breast with a wide mouth. It masticates the breast milk when it bites the areola and nipple and gobbles up the milk that gushes out. The movements of the jaws during masticating the milk are observable by the movement of both the temporal muscles and the ears (**Figure 1**).

At the start of breastfeeding the baby's pulse increases then decreases gradually and becomes constant. Its SpO_2 remains constantly high as determined by monitoring with a pulse oximeter. Sometimes the baby's forehead is moistened with sweat. The physiology of healthy breastfeeding is almost the same as that of slow jogging. There is good eye contact between the baby and the mother during breastfeeding.

In pathological breastfeeding, the baby sleeps and/or shuts its eyes during breastfeeding. It hardly latches onto the mother's breast. The masticating movements during feeding are hardly observable. No movements of temporal muscles and jaws are observable (**Figure 2**).

3.2. Studied Items with Regard to Babies' Breastfeeding

1. Whether the baby opens its eyes wide when latching onto the mother's breast. We counted it as negative when the eyes were half closed when suckling.
2. Whether the baby latches onto the mother's breast with its mouth wide open.
3. Whether a baby has a callus on its upper lip (**Figure 3**).

Figure 1. Healthy breastfeeding. Photo shown by consent of the mother.

Figure 2. Unhealthy breastfeeding. Photo shown by consent of the mother.

Figure 3. Callous on the upper lip.

Babies with ADEL have a short upper labial frenulum as well. The upper frenulum attaches to the upper labium and gingiva. The condition results in a narrow upper oral vestibulum.

When a baby latches onto the mother's breasts its upper lip turns inside and rubs against the skin of the mother's breast. As a result a relatively long and wide callus forms on the center of the upper lip (**Figure 3**).

4. Whether the baby moves its jaws during breastfeeding.

When a baby latches onto the breast the nipple is pushed into the sucking sulcus of the hard palate by the infant's tongue. Then the baby masticates the breast rhythmically with its jaws. Movements of both the temporal muscles and ears are visible when a baby masticates breast milk. When both movements were observed we counted it as positive. No movement and/or one movement were counted as negative.

5. Whether the baby has a white dorsum tongue by cornification (**Figure 4**).

Incorrect movements of the tongue during breastfeeding result in white cornification on the dorsum tongue. It causes pain and/or injuries to the mother's nipples.

6. Whether the baby makes sounds like "tsukon-tsukon" or "chupa-chupa", during breastfeeding. ("Tsukon-tsukon" or "chupa-chupa" is an onomatopoeia in Japanese. These are the suckling sounds that are heard as not soft but rather as hard clicking sounds.)

3.3. Mother's Breasts

Pathological breastfeeding affects both the mother's nipples and breasts.

(B) Changes in nipples after babies' surgeries were as follows:

7. Nipple pain.

8. Flattening of nipples (**Figure 5**).

Figure 4. White cornification on a baby's tongue.

Figure 5. Flattened nipple. The nipple is smashed flat from the areola to the top of the nipple. White erosion is observable on the top of the nipple.

9. White debris or cheese formation on top of nipples (**Figure 6**).
10. Presence of blisters on nipples.
11. Cracked nipples (**Figure 7**).

(C) Changes in the breasts after babies' surgeries

Healthy breasts of breastfeeding mothers are soft and appear healthy with no palpable masses within. The shapes of nipples, areola, and breasts are normal (**Figure 8** and **Figure 9**). The following conditions were examined before and after the babies' surgeries.

Figure 6. White debris. The areola is swollen with congestion.

Figure 7. Cracks on a nipple. Crusts are observable on the top. It can be said that the cracks were injuries from breastfeeding.

Figure 8. Healthy breast just after breastfeeding (side view). Healthy breast after breastfeeding is soft.

Figure 9. Healthy breast just after breastfeeding (frontal view).

12. Soft, swollen or shrunken breasts (**Figures 8-11**).
13. Pain after breastfeeding.
14. Presence of mastitis (**Figure 12**).
15. Presence of palpable masses.
16. If present, distribution of masses in the breasts.

A breast was supposed as a circle, a Cartesian coordinate was made at the center of a nipple, and the breast was divided into four quadrants. The right breast was divided into four quadrants counter-clockwise from the upper-ventral I quadrant to II, III and IV quadrants (**Figure 13(R)**). The left breast was divided the same way clockwise from the left upper-ventral I quadrant to the bottom ventral IV quadrant (**Figure 13(L)**).

Locations of palpable masses were noted within a quadrant. When a mass was palpable in both quadrants, it counted as being in both quadrants (**Figure 14**).

Contingency table by StatView was employed for statistical analyses.

Participants of this study were provided information on the procedures and purposes of the study before they were requested to provide written informed consent.

4. Results

(A) Suckling by babies

1. Whether the baby opens its eyes wide when latching onto the mother's breast.

Twenty-four percent of the babies (16 cases) opened their eyes and 76% (51 cases) shut their eyes during breastfeeding before the operations. One month after surgeries, 76% (51 cases) opened the eyes and 24% (16 cases) closed the eyes ($p < 0.0001$) (**Figure 15**).

2. Whether the baby latches onto the mother's breast with its mouth wide open.

Thirty percent of the babies (20 cases) latched onto the breasts correctly and 70% (47 cases) did not before the operations. One month after the surgeries, 84% (56 cases) did latch on correctly and 16% (11 cases) did not (**Figure 16**).

3. Whether a baby has a callus on its upper lip.

Seventy-three percent (49 cases) had calluses and 27% (18 cases) did not before operation. No callus in any baby was observed 1 month after surgeries ($p < 0.0001$) (**Figure 17**).

4. Whether a baby moves its jaws during breastfeeding.

Twenty-seven percent (18 cases) moved the jaws during breastfeeding and 73% (49 cases) did not before surgeries. One month after the surgeries, 88% (59 cases) moved their jaws and 12% (8 cases) did not during breastfeeding ($p < 0.0001$) (**Figure 18**).

5. Whether the baby has a white dorsum tongue by cornification (Photo 4).

Seventy percent of the babies (47 cases) presented with white debris and 30% (15 cases) did not before surgeries. One month after surgeries, 22% (15 cases) had white debris while 78% (52 cases) did not (**Figure 19**).

6. Whether the baby makes sounds like "tsukon-tsukon" or "chupa-chupa", during breastfeeding.

Forty-five percent of the babies (30 cases) made such sounds and 55% (37 cases) did not before the operations. However, 1 month after the surgeries 21% (14 cases) made such sounds and 79% (53 cases) did not ($p < 0.0055$) (**Figure 20**).

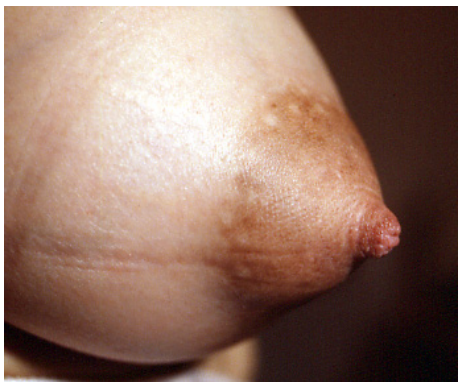

Figure 10. Swollen breast caused by congestion.

Figure 11. Shrunken breast, no function of the mammary glands is observable. Breast is shrunken and hangs down.

Figure 12. Mastitis (left). Both breasts are swollen with congestion.

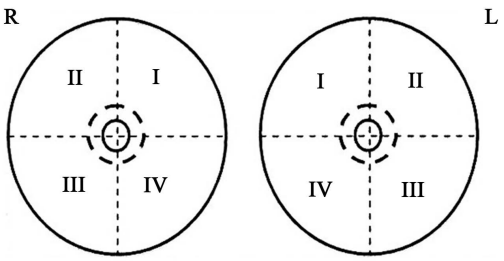

Figure 13. Cartesian coordinate of breasts. Right breast was counted counter-clockwise from the upper-ventral I quadrant to the II, III, and IV quadrants. The left breast was **counted** clockwise from the left upper-ventral I quadrant to the bottom ventral IV quadrant.

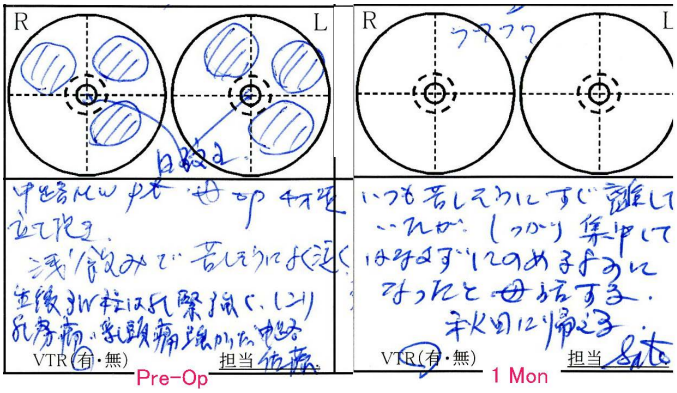

Figure 14. An example of the distribution of palpable masses from a research paper before and 1 month after surgeries from a mother of a 1-month-old boy. Before surgeries (Pre-Op). Palpable masses were identified in the I, II, IV quadrants in the right breast and in the I, II, III quadrants in the left breast. White debris was observed in both nipples. In the reference, it was written that the mother had undergone CGL and EVO when she was 4 years old. She held her baby in a standing position. When breastfeeding, his suckling was very shallow. He cried frequently with suffocating sounds. From his birth to 3 weeks the mother had intense nipple and breast pains when breastfeeding. (Signature; Sato in Japanese) 1 month after surgeries (1 mo); both breasts are soft. In the reference, it was stated that the mother said, "He looked as if he was suffocating and he did not continue breastfeeding before surgeries. Now he concentrates on the breastfeeding and continues until he is finished." They returned to their home in Akita city in the northern part of Japan. (Signature; Sato).

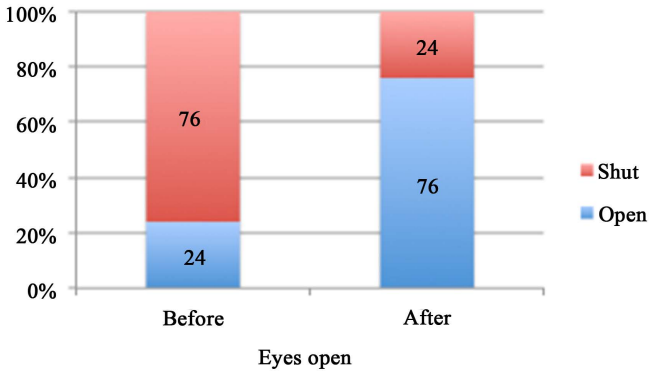

Figure 15. Whether the baby opens its eyes wide when latching onto the mother's breast ($p < 0.0001$).

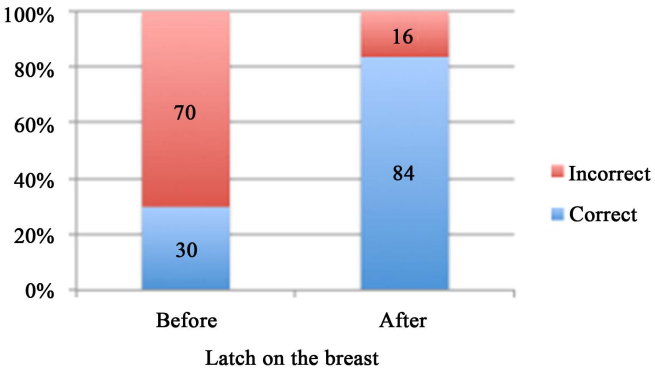

Figure 16. Whether the baby latches onto mother's breast with its mouth open wide ($p < 0.0001$).

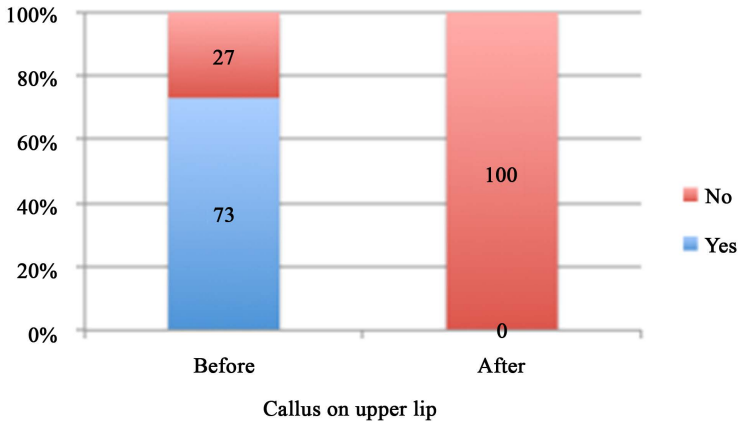

Figure 17. Whether a baby has a callus on the upper lip ($p < 0.0001$).

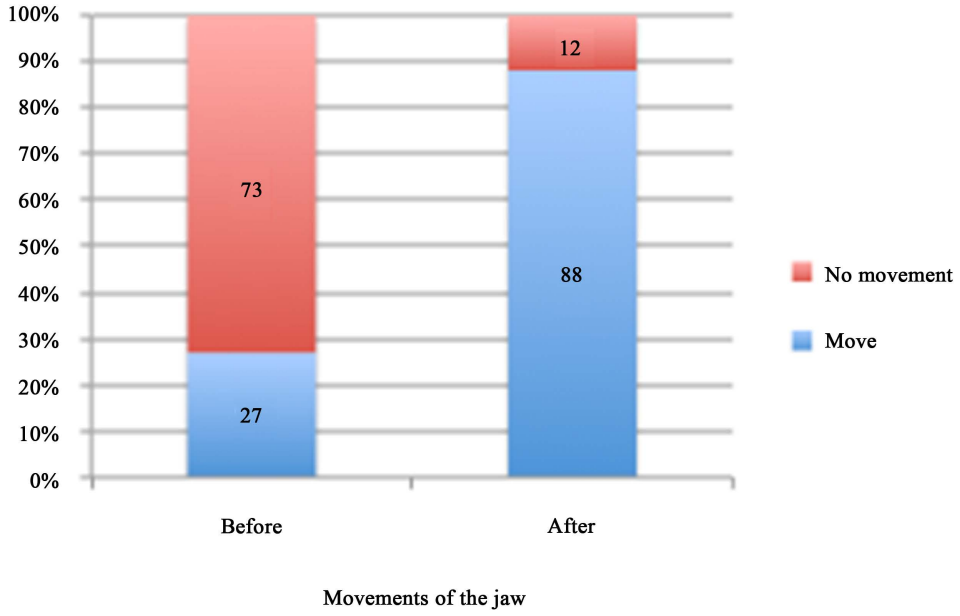

Figure 18. Whether the baby moves its jaws during breastfeeding ($p < 0.0001$).

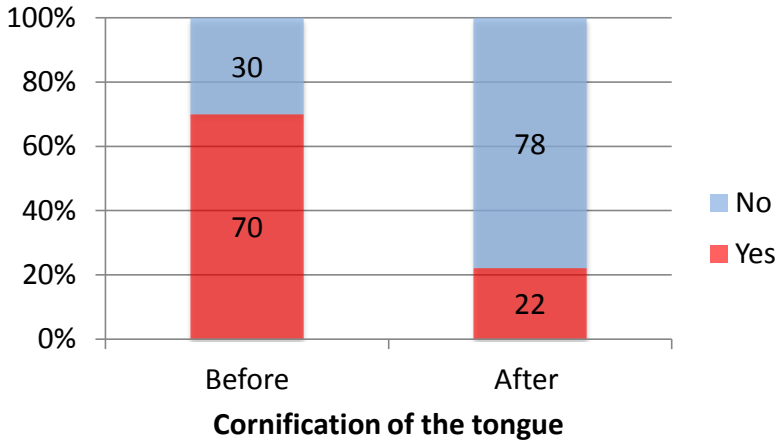

Figure 19. Whether the baby has white dorsum tongue by cornification ($p < 0.0001$).

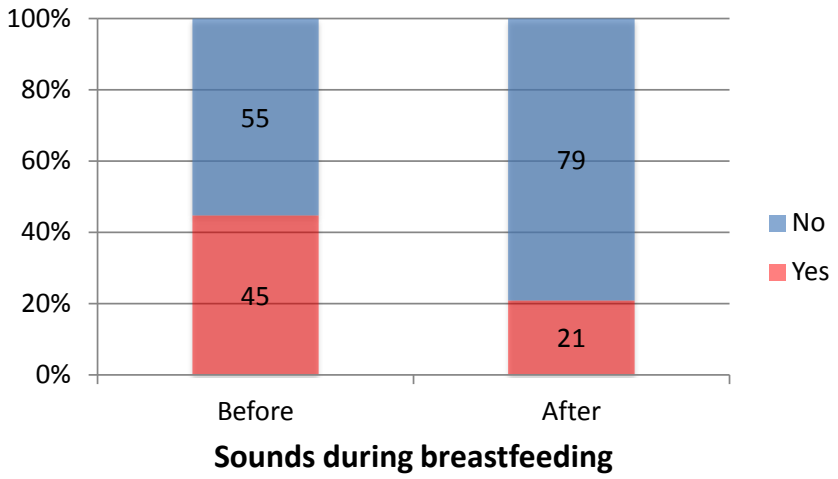

Figure 20. Whether the baby makes sounds like "tsukon-tsukon" or "chupa-chupa", during breastfeeding ($p < 0.0055$).

(B) Mothers' nipples

7. Nipple pain.

Of the mothers, 67% (45 cases) had nipple pain before the babies' surgeries and 6% (4 cases) had the same degree of pain 1 month after the operations ($p < 0.0001$) (**Figure 21**).

8. Flattening of nipples.

Before the babies' operations, 55% (37 cases) of the mothers had flattened nipples but only 7% (5 cases) had the same condition 1 month after the operations ($p < 0.0001$) (**Figure 22**).

9. White debris or cheese formation on top of nipples.

Before the babies' operations, 39% (26 cases) of the mothers had white debris on the nipples and only 7% (5 cases) had this condition 1 month after the operations ($p < 0.0001$) (**Figure 23**).

10. Presence of a blister on nipples.

Of the mothers, 3% (2 cases) had blisters before the babies' operation and the same 3% (2 cases) had blisters on their nipples 1 month after the operations (**Figure 24**) ($p > 0.9999$). Both mothers fed their babies by mixed feeding, with bottle feeding predominant. Nipples of both mothers were short.

11. Cracked nipples ($p > 0.9999$).

Only one mother had cracked nipples (**Figure 25**). Before her baby's operations both of her nipples had cracks but 1 month after the operations only the left nipple had a crack. Her baby fed well from the right breast but clumsily from the left breast.

(C) Mothers' breasts

12. Soft, swollen or shrunken breasts.

Before the babies' operations, breasts were soft in 48% (32 cases), swollen in 43% (29 cases), and shrunken in 9% (6 cases) of the mothers, and 1 month after the babies' surgeries breasts were soft in 90% (60 cases), swollen in 6% (4 cases), and shrunken in 4% (3 cases) of the mothers (**Figure 26**). There was a significant difference between before and after surgeries ($p < 0.0001$).

Five cases that had shrunken breasts before the babies' operations had soft breasts after the operations and one had shrunken breasts both before and after the operations. Two of three mothers who had shrunken breasts after the babies' surgeries had soft breasts before the operations. These two mothers changed from breastfeeding to bottle feeding. The mother that had shrunken breasts before and after the baby's operation had provided her baby with breast milk obtained by a breast pump before the surgeries. After the surgeries, she changed to bottle feeding.

13. Pain after breastfeeding.

Twenty-three percent (13 cases) of the mothers had breast pain before the babies' surgeries and 4% (3 cases) had breast pain 1 month after the operations ($p < 0.0031$) (**Figure 27**).

14. Presence of mastitis.

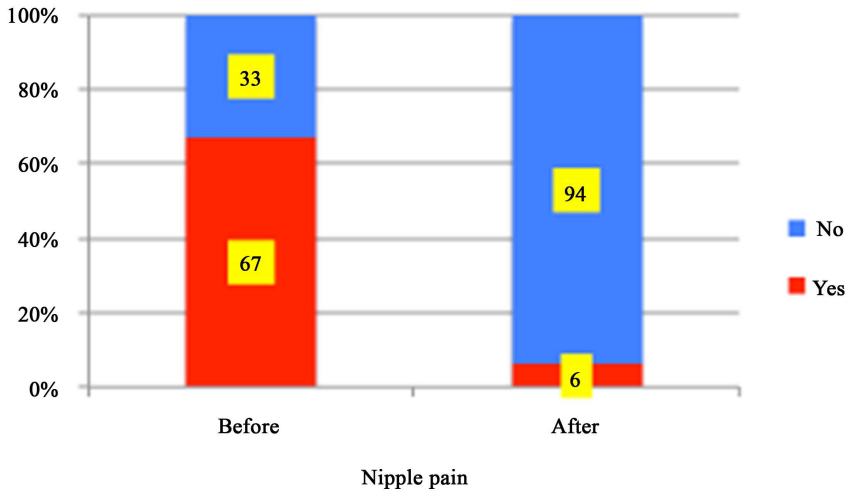

Figure 21. Nipple pain.

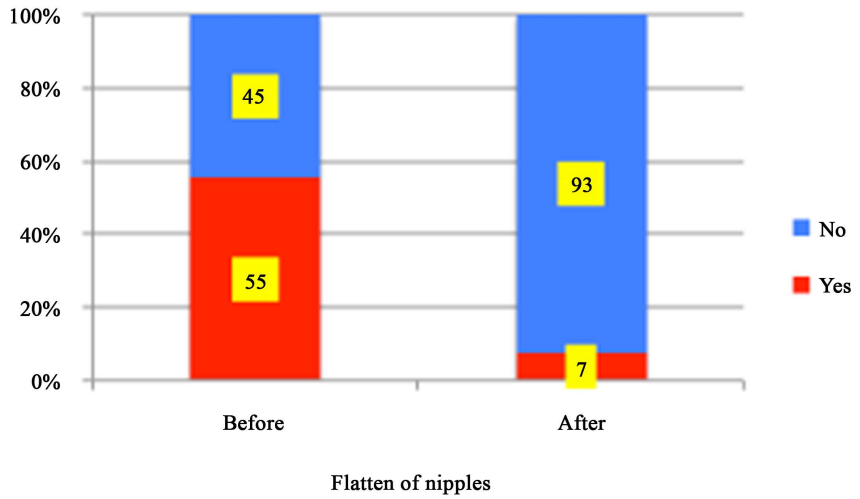

Figure 22. Flattening of nipples.

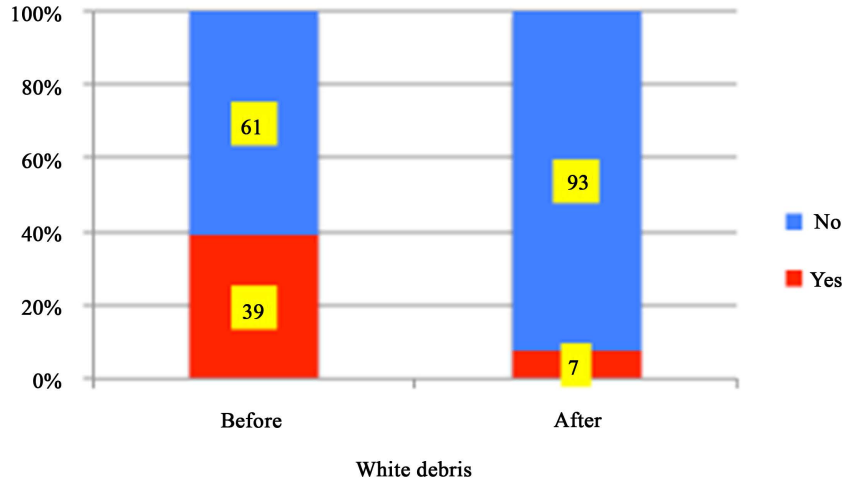

Figure 23. White debris or cheese formation on the top of the nipples.

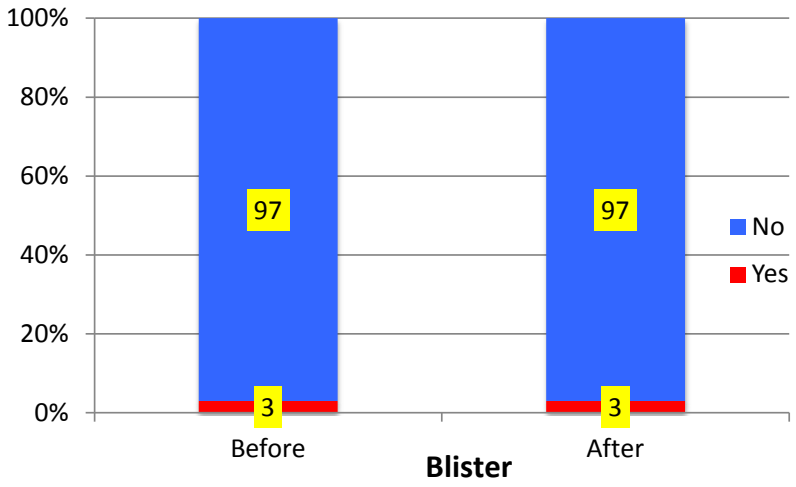

Figure 24. Presence of a blister on **a** nipple.

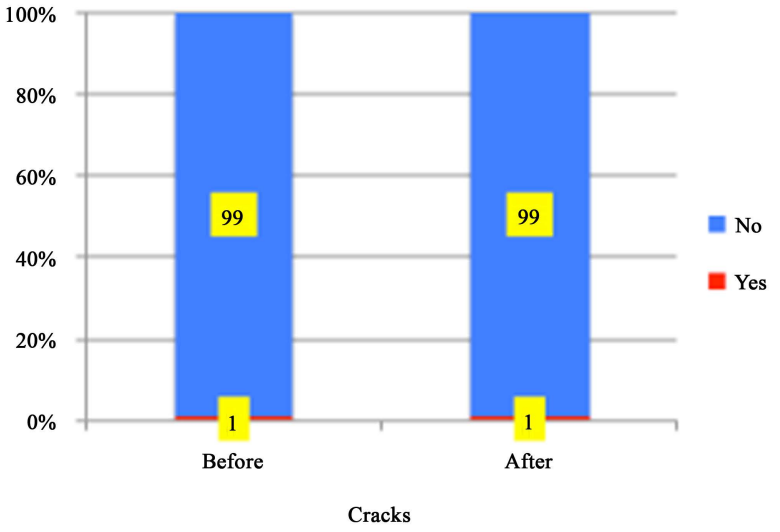

Figure 25. Cracked nipples.

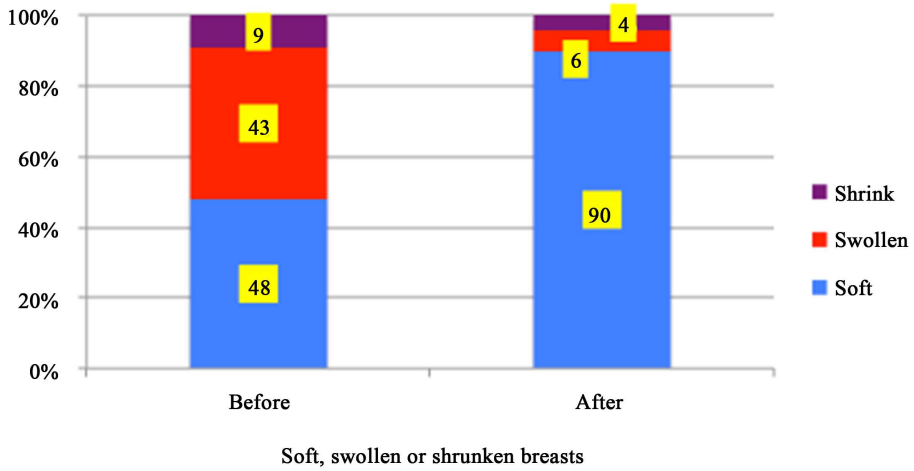

Figure 26. Soft, swollen or shrunken breast.

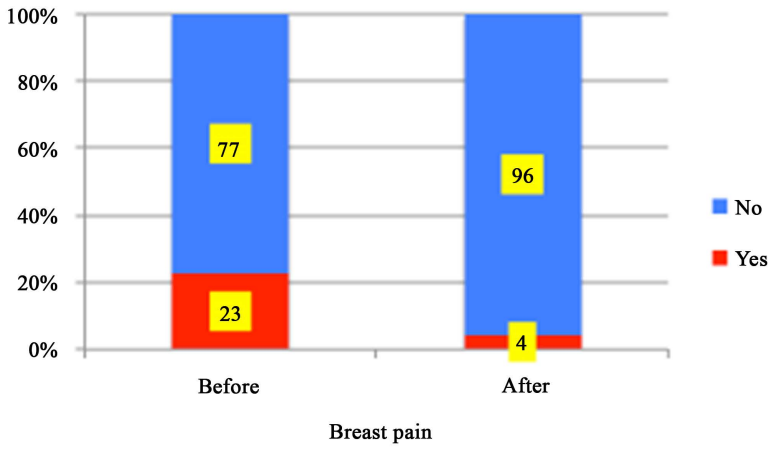

Figure 27. Pain after breastfeeding ($p < 0.0031$).

Nine percent (6 cases) of the mothers had mastitis before the babies' operations and 1% (1 case) had mastitis 1 month after the operations (**Figure 28**) ($p = 0.0174$). There was a significant difference in the prevalence of mastitis before and after the babies' surgeries ($p < 0.05$).

15. Presence of palpable masses in a breast. When a mass was palpable in a breast that breast was counted as positive for a mass.

Of the mothers, 73% (49 cases) had palpable masses before the babies' operations, but masses were evident in only 25% (17 cases) 1 month after the surgeries. The size of these masses that remained in the same place as before the operations had diminished 1 month after surgeries ($p < 0.0001$) (**Figure 29**).

16. If present, distribution of masses in the breasts.

In the right breasts, there were 105 masses and 80 masses in the left breasts. One month after the babies' operations there were 10 masses in the right breast and 30 masses in the left breast (**Figure 30**).

Before the surgeries, distribution of masses in each quadrant in the right breast was as follows: 27 (26%) in the I right quadrant (RQ), 42 (40%) in the II RQ, 26 (25%) in the III RQ, and 10 (9%) in the IV RQ. In the left breast there were 21 masses (26%) in the I left quadrant (LQ), 36 (45%) in the II LQ, 14 (17%) in the III LQ, and 9 (11%) in the IV LQ (**Figure 30** up, Pre-Op, red).

Although the right breasts had more masses (105) than the left breasts (80), distribution rates of the masses in each quadrant were almost the same in the right and left breasts. Masses were most frequent in the II quadrant of both breasts. Next to the II, frequencies were as follows: I, III, and IV quadrants. (**Figure 30**, above) (Pre-Op, red). The rates of distribution of masses in the left breasts were almost the same before and after the babies' operations (Fi. 31, below) (1 m, blue).

5. Discussion

(A) Suckling of babies

Healthy breastfeeding and pathological breastfeeding are described above. There are some lasting results of pathological breastfeeding. For example, the lack of mastication during breastfeeding leads to poor development of both the mastoid and mandibular bones, poor dental arches, misaligned teeth, and malocclusion. The baby's mouth, nostrils, and nasal cavities become narrow as well. Lack of jaw movements leads to accumulation of cerumen.

During pathological breastfeeding, the baby's heart rate is high and SpO_2 unstable. In some babies, the SpO_2 falls below 80 during pathological breastfeeding. Frequently feeding cannot continue due to suffocation and sometimes vomiting. The SpO_2 goes down during these episodes, which might be caused by deviation of the epiglottis and larynx. Body temperature is also lower than normal.

With normal breastfeeding the epiglottis and larynx move upward and connect with the choanae. The baby can breathe while breastfeeding. In most babies with ADEL the epiglottis deviates right and forward. The larynx deviates up and forward. Sometime it bends far forward and the ostium of the esophagus is visible. In these conditions the connection between the choanae and the larynx is not smooth, and suffocation and vomiting could

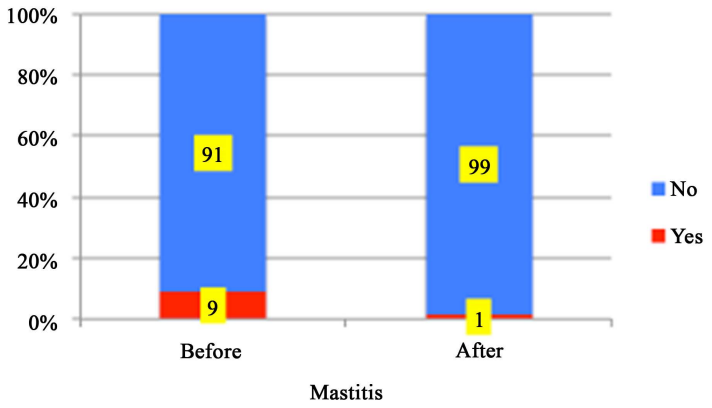

Figure 28. Presence of mastitis ($p < 0.05$).

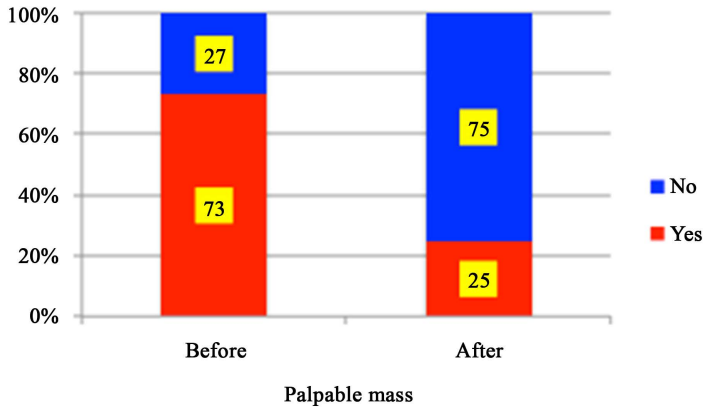

Figure 29. Palpable masses ($p < 0.0001$).

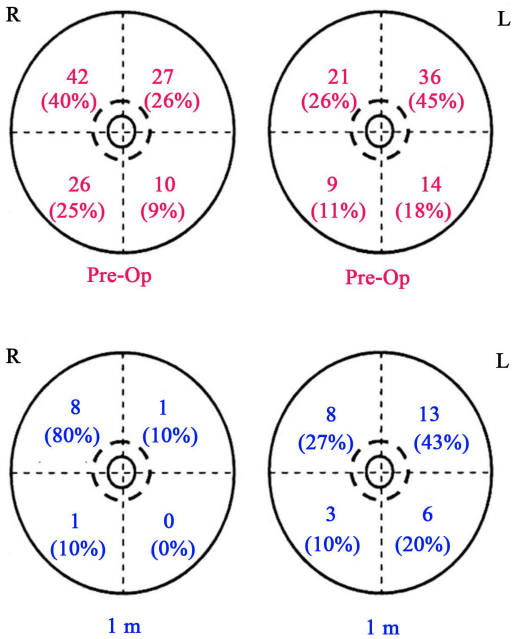

Figure 30. Distribution of masses in the breasts. (Before operations: Pre-Op in red. One month after operations: 1 mo in blue.)

occur during breastfeeding. In addition, the baby's head is distorted in the direction of the deviation of the epiglottis and larynx. To make respiration easier the baby turns its face toward the direction of the laryngeal deviation [21]. This leads to distortion of the body and has resulted in lumbago in the aged [22] [23].

The rate (73%) of callus on the upper lip of the babies was high. Not a small portion of babies with ADEL have a short upper lingual frenulum, which restricts the upper lip and gingiva with the result that the upper oral vestibulum is narrow. When a baby bites the mother's breasts its upper lip turns inside and the lip rubs against the skin of the mother's breast. As described above, a baby with ADEL does not fully open its mouth. Both result in a relatively long and wide callus on the center of baby's upper lip. In addition, the narrow vestibulum restricts the development of the maxillary bone as well as facial muscles.

The EVO is a surgery that cuts the upper frenulum and detaches the depressor septi nasi (DSN) muscles from the maxillary bone. Lateral growth of the maxillary bone is accelerated by these procedures. Both expansion of the nose and nostrils are observed after EVO. In addition, development of facial muscles that is restricted by DSN muscles can proceed and the mouth and eyes widen [13] [15] [24] [25].

The decrease in the percentage of callus (0%) after surgeries indicated that the baby could latch onto the breast well after EVO and CGL. Widening of the upper vestibulum and opening of the mouth were easier after these procedures.

We performed research on 5-year-old children who did and did not undergo EVO and CGL at under 4 months of age. The results showed that occlusions and alignment of the teeth of the operated group were far better than among those in the non-operated group. In addition, no child had nasal septal deviation in the operated group. These studies revealed that healthy breastfeeding is mandatory for healthy development [26]-[28].

The cornification of the dorsum tongue and making sounds when feeding mean that movements of the tongue are not smooth but are unnatural.

A baby has a sulcus in its hard palate. When breastfeeding, the baby latches onto the breast and the nipple comes in contact with the sulcus. The baby begins feeding with its tongue moving peristaltically [28]. If this feeding is normal no sound is audible. When the mother's nipple does not latch on to the sulcus of the hard palate, suckling movements make "tsukon-tsukon or chupa-chupa" sounds. This manner of sucking causes aerophagia, belching and/or flatus as well as abdominal swelling. The dorsum tongue causes pain in the mother's nipple.

Mothers

Damage by breastfeeding is mainly to the mother's nipples and breasts.

(B) Nipples

Nipple pain (67%), flattening of the nipples (55%), and white debris (39%) were observed in many nipples before the babies' surgeries. Thereafter they were markedly decreased ($p < 0.0001$). It is obvious that these conditions resulted from incorrect feeding by the babies.

As seen in babies with ADEL, rate of latching onto the breast was low at 30% and callus on the upper lip was high at 73% as were cornification of the dorsum tongue (70%) and no movements of the jaw (73%). These results suggested that the babies did not effectively latch onto the breast, resulting in shallow breastfeeding. Scant movement of the jaw means that the baby is mainly sucking the nipple. Its cheeks become hypertrophic and the temporal regions are concave because of the predominant suckling movement rather than mastication. In addition, both the high rate of cornification of the dorsum tongue and callous on the lip suggest that the baby rubs and bites the nipple. These cause pain and flattening of the nipple. This improper breastfeeding causes nipple debris. When this kind of feeding is too strong, the nipple is naturally hurt and blistered.

Two cases of blisters (3%) and one of cracks (1%) were observed. Both were evident in these mothers before and after the infants' surgeries. In the case with cracked nipples, both nipples were cracked before surgeries and only one after surgeries. The two mothers with blistered nipples mainly bottle fed their infants because their nipples were abnormally shaped. In addition, their babies were not skilled in taking the nipples into their mouth**.** These facts suggest that breastfeeding requires practice by both infants and mothers.

(C) Mothers' breasts

In this study, palpable masses (73%), swollen breasts (48%), breast pain (23%), and mastitis (9%) were identified.

Nipple pain appeared mainly when breastfeeding while breast pain appeared mainly after nursing. This pain might be caused because the strength of suckling is uneven regarding the mammary glands. Imbalance of the strength exerted on mammary glands causes inflammation of the glands. This means that the breast pain might

be secondary to abnormal breastfeeding.

The rate of mastitis was high (9%) before and low (1%) after the babies' surgeries, showing a significant decrease ($p < 0.05$). The cause of mastitis might be the same as the cause of breast pain and palpable masses.

The ratio of palpable masses (73%) was very high and might have been caused by some problems with the mammary glands, such as congestion and/or inflammation. Although the number of palpable masses was greater in the right breast, the rates of palpable masses in each quadrant of both breasts were almost same. The highest number of masses were in the II quadrant in both breasts, followed by those in I quadrant. Masses in III and IV quadrants were fewer than in I and II. Palpable masses mainly appeared in the upper half of the breasts. These appearances of palpable masses indicate that the pathology is the same in both breasts.

When breastfeeding, the upper jaw or maxillary is on the upper breast in I or II and the lower jaw or mandible is on the lower breast in III or IV. The lower jaw or mandible continues pressing the lower breast and upper jaw or maxillary does not move but continues to receive the strength of lower movements. Breastfeeding involves repetitive movements with a pumping action that forces out milk in the mammary ducts.

When there are some deviations in strength of upper and lower movements during latching on, not only do masses accumulate in the breast, but also pain results after breastfeeding. These deviations or imperfections in latching on might be the cause for the higher rate of palpable masses in the upper half than the lower one. It also suggests that lower movements have a kind of massage action on the breasts.

Calluses on the upper lips of babies as well as the rate of no movement of jaws of over 70% might prove deviations and imperfections in latching on. It can be said that no masses in the breasts would develop when the movement is correct.

Palpable masses after surgeries decreased from 105 to 10 in the right breast and from 85 to 30 in the left breast. The amelioration rate in the right breast was 90.5% but in the left breast was only 62.5%. Healing of the left breast was retarded obviously. The causes of the retardation in healing might be that a multitude of people are right handed as well as the fact that the right deviation of the epiglottis and larynx is more numerous in ADEL. In addition, babies might be able to latch onto the right breast easier than the left breast [21]-[23].

I tasted breast milk directly from a healthy breast. The milk was very smooth, and tasted neither salty, nor sour, nor bitter, nor sugary, nor unsavory. It was extremely delicious and moving. It was absorbed unaffectedly in the mouth. It manifested motherhood. The moment I tasted it, I understood that the baby that nurses from a healthy breast is happy.

6. Summary

Babies that are born with ADEL cannot breastfeed correctly and hurt their mothers' breasts. ADEL not only hinders the normal development of upper and lower jaw bones, nose, and occlusions, but also prevents mothers from breastfeeding. It prevents communication between mother and child as well as others.

Remarkable ameliorations of these reported cases, both of babies and mothers, revealed that surgical interventions are necessary for babies with ADEL. In addition, it proved that aid in breastfeeding by midwives is invaluable for mothers.

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