Three case reports of infants with ankyloglossia affecting breastfeeding

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SUMMARY

Ankyloglossia is a congenital anomaly which may reduce or restrict the tongue tip mobility. The restricted mobility is caused by an unusual short, thick lingual frenum. This condition may cause various problems in infants including breastfeeding in the new-borns. This case report describes 3 cases of ankyloglossia affecting breastfeeding and highlights the experiences of the mothers and their difficulties in breastfeeding babies with it. Comprehensive feeding examination was accomplished, the primary cause of feeding issues was identified, and frenotomy intervention was provided. Post frenotomy, infants were able to breastfeed easily and this was beneficial in continuation of breastfeeding and pain reduction in mothers.

INTRODUCTION

The tongue is an important organ; not only for development of speech but also for dental occlusion determination, nutrition and swallowing.[1] The tongue moves freely and is attached by a lingual frenulum to the floor of the mouth, a fibro-mucosal fold that connects the ventral surface of the tongue and the mucosa of the oral pavement.[1] At birth, the connecting tissue regressed and disappeared. However, when it fails to regress, this tissue remnant may interfere with tongue mobility, known as 'ankyloglossia' or tongue-tie.

The prevalence of ankyloglossia varies from 4% to 10% depending on the diagnostic criteria used.[1] It is said that the incidence of ankyloglossia is approximately 0.2% to 5% depending on the population studied and more common in males.[1] It has been associated with other syndromic conditions such as Smith-Lesli-Opitz– syndrome, Beckwith Wiedman Syndrome and orofacial digital syndrome. These cases illustrate the management of ankyloglossia of 3 infants who had breastfeeding difficulties. The treatment objectives were to achieve successful breastfeeding for both mothers and infants as well as to eliminate the nipple pain.

Case 1

A 3-month-old girl was brought to the lactation clinic with a complaint of breastfeeding difficulties and seeking a second opinion. Her mother was distressed as on each feeding session due to difficulty in latching since birth and was very keen in continuing to breastfeed her child. The infant was diagnosed with ankyloglossia at birth but was given appointment for one year later for review which caused the mother to start formula feeding. She had no breastfeeding difficulties with her other two older children and was able to breastfeed them successfully.

On examination, the infant was alert, healthy and developmentally appropriate. Even though the breast appeared normal with no breast or nipple trauma or surgery, breastfeeding observation showed poor attachment. Correct way of attachment and positioning for a successful breastfeeding was taught by a lactation specialist. Unfortunately, she was still unable to sustain her latch. Intraoral examination showed that there was ankyloglossia class 2 (Figure 1). Hazelbaker Assesment Tool for Lingual Frenulum Function (HATLFF) score was 5 for appearance and 7 for function. She was subsequently referred to a paediatric dentist for frenotomy.

Evaluation by the paediatric dentist noted that the tongue movement was restricted in three planes where elevation, lateral and extension motion were reduced. Frenotomy was suggested and the mother consented. Local anaesthesia (LA) applied to the infant's frenulum. The tongue was carefully lifted using a sterile grooved retractor while the infant was appropriately positioned and restrained. The attached frenulum was snipped using a sterile Iris scissors. Care is taken to avoid injury to any other adjacent tissues and structures. Very minimal bleeding occurred and pressure using sterile gauze was applied to the frenotomy site.

Immediately post frenotomy, the infant was able to latch well. Free tongue movement was achieved and the HATLFF score improved to 10 and 13 respectively when reviewed the next day. Post-operative intraoral stretching exercises was advised to be done for 3 weeks. For the intraoral exercise, the mother is advised to put her finger under the tongue and pushing into the centre of the wound, moving side by side. Both cheek and palate also need to be stimulated by a finger. Four weeks post operation, breastfeeding continued well, and the mother was happy with the outcome.

Case 2

A 6-week-old boy was brought for breastfeeding evaluation. He was the first-born in the family. His mother complained of nipple pain on each feeding session with pain score of 7/10 since birth. The home visit nurses helped with correcting the position and attachment but still the mother experienced nipple pain while feeding. Despite the pain, the mother continued expressing her milk and feed the infant via bottle.

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Fig. 1: Ankyloglossia Class 2 and appearance post frenotomy

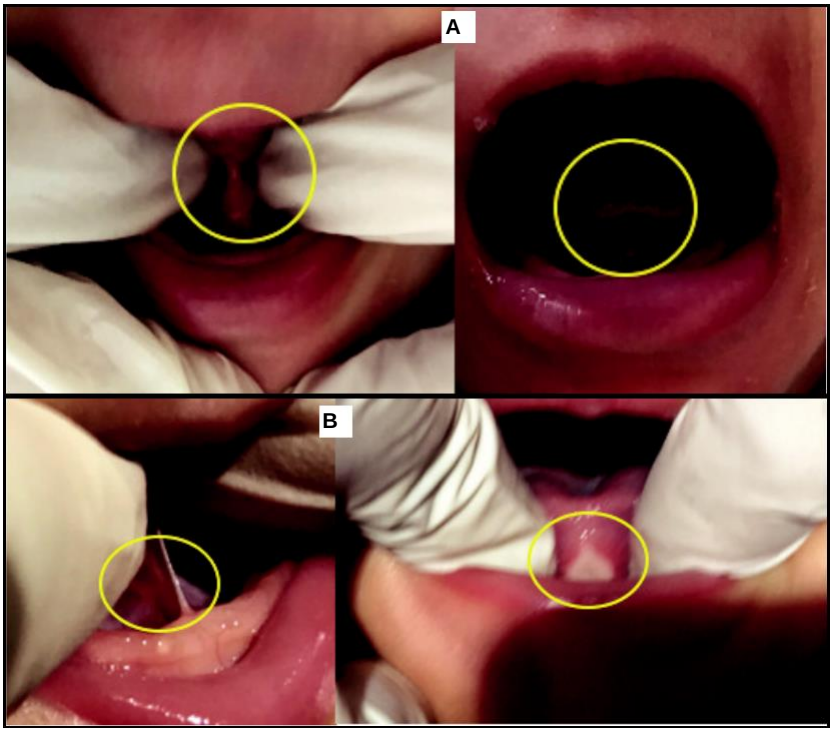

Fig. 2: a) Ankyloglossia Class 3 and appearance post frenotomy. b) Ankyloglossia Class 2 and appearance post frenotomy

On examination, the infant was alert and his anthropometry measurements were satisfactory. Breastfeeding observation showed that he had poor attachment and could not sustain his latch. Intraoral examination revealed ankyloglossia class 3 (Figure 2). Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) score for appearance was 8 and 9 for function. Other systemic examinations were unremarkable. The breast examination of the mother was found to be normal. The infant was referred to a paediatric dentist for frenotomy.

Frenotomy was done under LA similar to Case 1 with post oral stretching exercise advice given. His latch improved and the pain score of the mother's reduced tremendously. The HATLFF score improved to 10 and 14.

Case 3

A 15-day–old boy presented with poor sucking and difficulty in breastfeeding since birth. He took a longer time to complete his feed compared to previous siblings. His mother also had painful and cracked nipple with a pain score of 8/10.

On examination, the infant was jaundiced. His weight was 2.5kg. Breastfeeding observation showed that he had poor attachment and would come off the breast easily. Intraoral examination showed ankyloglossia class 2 (Figure 3). HATLFF score was 8 and 7 respectively. Other systemic examinations were unremarkable. Breast examination of the mother revealed engorged breast with right nipple abrasion. Despite correction of attachment, he too was referred for frenotomy.

Frenotomy was done similar to the other two cases. His mother reported instant reduction of pain and he was sucking effectively. Post frenotomy, HATLFF score improved to 9 and 10. Post-operative oral stretching exercise was advised. Four weeks post frenotomy, he continued breastfeeding and thriving well.

DISCUSSION

Ankyloglossia has been associated with breastfeeding difficulty, which can cause cessation of breastfeeding before the recommended time. In newborn babies, difficulty in

breastfeeding can range from poor weight gain to refusing the breast.[2] 25% of mothers with ankyloglossia infant had feeding difficulty and nipple pain.[2] As in this case series, all mothers had difficulty in latching the infant and two mothers complained of nipple pain.

Ankyloglossia usually goes unnoticed. Symptoms such as poor latching, infant slipping off the breast while feeding, severe nipple pain, incomplete breast drainage and poor weight gain were the most common presenting complaints.[3] All infants in this case series had problems whereby they easily slipped off from the breast while feeding.

Recognising ankyloglossia can be a challenge to primary care physicians as the criteria for diagnosis and classification still remain unsolved.[4] Ankyloglossia classification can be assessed by the tongue's anatomy and functional status such as using the Kotlow's classification and HATLFF.[5] In Kotlow's classification, it consists of descriptions of appearance of both tongue and frenulum with its absolute measurement.

Meanwhile, HATLFF is a semi-quantified scoring system used to classify severity of ankyloglossia by lactation consultants. It covers both the appearance as well as the functional status. The shape of the tip of tongue, the elasticity and length of the frenulum and its attachments at the tip of the tongue and the floor of the mouth is assessed in appearance score. Lateralisation, the lift and protrusion of the tongue, cupping and snapback are ascertained in the functional status. It has been shown to be highly correlated with difficulty with latching the infant onto the breast as well as sore nipple experienced by mothers.[5,6] For HATLFF, a total score of 8 or less for appearance and total function score of 11 or less is considered as severe ankyloglossia and significant for division as seen in our cases.

There are many other tools developed to assess the severity of ankyloglossia and decision for frenotomy which are Coryllos Classification, Bristol Tongue Assessment Tool (BTAT) and Frenotomy Decision Tool for Breastfeeding Dyads (FDTBD) assessment.[5] For family physicians without further training in lactation, the most useful assessment is based on the position of the frenulum attachment to the tongue base and degree of maximal tongue protrusion. In all of our cases, both Kotlow's criteria and HATLLF were used as the physician is more familiar.

The management of ankyloglossia is still debatable. There is no clear consensus on the indication of performing frenotomy and the timing of division.[5] Some researchers advocate a wait and see approach as in some children, the tongue movement can be compensated by them or in some milder forms may resolve with growth. However, there is no biomechanics study on the functional anatomy of the lingual frenulum to answer that the tongue tie loosens over period of time. Therefore, it would be recommended to examine the function of the tongue and lingual frenulum at the time the concern is raised.[1,5]

Based on the available evidence and since there is no absolute association between ankyloglossia and breastfeeding, not all babies with breastfeeding problems requires surgical division.[5,6] However, a thorough intraoral examination and breastfeeding assessment should be performed in babies with feeding difficulties by an International Board-Certified Lactation Consultant (IBCLC) to examine associated difficulties, counselled on proper technique, positioning and attachment of breastfeeding. The proper examination is essential as it will determine the etiology of the breastfeeding problem and to suggest further treatment effectively.

A review article in 2018 suggested that the only relevant criteria for frenotomy is medical breastfeeding related problem which often present during the neonatal period and early infancy.[5] The Academy of Breastfeeding Medicine (ABM) also recommends that infants with ankyloglossia having significant breastfeeding difficulties after thorough evaluation may undergo frenotomy.[6]

Based on the clinical score and symptoms experienced by the mother-infant dyad, frenotomy was performed in all three cases. Frenotomy is a simple procedure which results in improvement of breastfeeding as suggested in recent reports.[3-5] Post frenotomy, most infants are able to feed immediately without any problem with significant reduction of pain in mothers.[3]

After frenotomy, the wound heals by secondary intention instead of primary closure. Extraoral and intraoral exercises are recommended to ensure that the tissue remains free from restriction and prevent reoccurrence. Rotational movement over the inner aspect of the cheeks and palate during intraoral exercise will help in improving the sucking reflex of the babies.[3]

CONCLUSION

These case reports highlight the importance of recognising ankyloglossia in babies with significant breastfeeding difficulty by primary care physicians. A thorough and careful evaluation of the intraoral cavity and breastfeeding assessment is recommended. Ankyloglossia with significant breastfeeding difficulties may warrant a frenotomy. Frenotomy is safe and successful in improving breastfeeding difficulties and reducing nipple pain for all the three cases.

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