Clinical Case Report

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Therapeutic effect of a cleft lip teat on infants with respiratory and feeding disorders

Two case reports

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Abstract

Rationale: Existing research into the effects of teat application has mainly focused on its negative and positive influence on the development of the oral cavity. Our work demonstrates that apart from changing the setting of the articulatory organs, the teat can also affect the quality of breathing, eating and sleeping.

Patients concerns: We described the cases of 2 children: a 19-month-old girl and a 2.5-month-old boy, who had breathing disorders due to withdrawal of the tongue and impaired food intake.

Intervention: The babies were bottled fed with a special teat for cleft lip patients to observe the influence of the teat on the setting of the articulatory organs and breathing.

Diagnosis: We suspected that the specific construction of the teat—the wide outer part and the short internal part—would affect children's reflexes and articulatory organs so as to force the frontal position of the tongue, which was meant to facilitate breathing and eating.

Outcomes: It was found that feeding with the cleft lip teat stimulates the gyro-linguistic muscle, which results in the proper position of the tongue and consequently better breathing and improved quality of sleep.

Lessons: A specialist bottle teat designed for babies with cleft lips can constitute an effective tool in the therapy of nonspecific respiratory disorders resulting from improper position of the tongue and other articulatory organs.

Abbreviations: CNS = central nervous system, MRI = magnetic resonance imaging, NREM = non-rapid eye movement, OSA = obstructive sleep apnea, PUM = Pomeranian Medical University, SIDS = sudden infant death syndrome, UA = upper airways, WHO = World Health Organization, WNL = within normal limits.

Keywords: articulatory movement, speech-language therapy, sucking, teat

1. Introduction

According to the World Health Organization (WHO) guidelines breastfeeding is the only feeding method that ensures proper, harmonious development of an infant and meets the standards for appropriate care of the newborn.[1] Sucking is critically important for newly born infants and young babies—it allows unassisted nursing and contributes to full development of the speech organs. The ability to suck results in better digestion, development of behavioral functions and prevents aspiration.[2]

Natural breastfeeding is not always possible, though, and a lot of newborns cannot do without bottle teats. Despite the widespread use of them in Western countries (75%–85% of children),[3] both pacifiers and bottle teats often become the subject of heated debates among paediatricians and parents promoting nursing as the best feeding method that ensures newly born infants and young babies their proper development.[4]

It has been showed that using a bottle nipple for feeding or a pacifier soothing a baby brings about a considerable number of negative effects. Prolonged use of a pacifier has been found to disturb the sucking reflex.[5] Other studies have demonstrated correlation between using a pacifier and termination of breastfeeding before a baby reaches 6 months of age.[5] Also, it has been found that prolonged use of a pacifier (children aged 6 months–2 years) results in an increased risk of otitis media or malocclusion.[6] Other studies show that sucking a pacifier may cause choking, food poisoning, development of allergies, increased risk of tooth decay as well as intestinal parasitic infection.[5] Latest studies, however, indicate that the use of a bottle teat is a sign of mothers' difficulties breastfeeding or lowered motivation for this mode of feeding rather than the root cause of early weaning.[7]

In the meantime, there are increasingly more scientific publications describing the benefits of a pacifier.[1] One of the

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most frequently quoted arguments for the use of a pacifier is its effectiveness in soothing premature and sick babies.[1] What is more, pacifiers may be used as a training to prepare prematurely born infants to natural feeding.[8] Pacifiers have also been shown to have a positive effect on the development of premature babies by both stimulating the sucking reflex and enhancing motor development of the articulatory organs.[9] Some studies have also demonstrated that babies who use pacifiers produce better physiological parameters—they are better oxygenated and have lower heart rate.[10]

Sucking stimulation was also found to affect blood glucose concentration as sucking stimulates the vagus nerve (nerve X), which is responsible for lowering the level of somatostatin and the secretion of gastrin.[10] It allows children to calm down, lengthens sleep, and consequently reduces energy consumption.[11] It was also determined that a pacifier increases a baby's alertness during feeding, which results in better food intake.[12] It improves the coordination of sucking, swallowing and breathing.[13] Also, a pacifier has a stimulating influence on the development of the central nervous system (CNS), maturation of oral reflexes and reduced pain perception.[11] The greatest benefit of pacifier use in newborns and young babies, though, is that it protects them from sudden infant death (SIDS).[14]

The above mentioned benefits of a pacifier and a bottle teat mean a faster gain of body mass and shorter length of hospital stay.[4]

This work aims to assess breathing and sleep disorders in relation to the structure and functioning of the articulatory organs in infants and discuss, on the basis of the presented cases, progress in the neurological speech therapy with a cleft lip bottle teat administered to young babies afflicted with disturbed breathing, sleep and food intake.

2. Methods

The authors received approval of Research Ethics Committee of PUM in Szczecin on March 15, 2018; No. of the approval: KB-0012/145/03/18. A written informed consent for the publication of the case was obtained from all of the subjects' parents or legal guardians.

3. Case reports

3.1. Case presentation 1

Subject: currently 19-month-old baby girl, born with cerebral palsy in pregnancy II, delivery II through an emergency cesarean section (suspected placenta abruption) at a gestational age of 26 weeks, birth weight 500 g, the Apgar score: 0/0/3/3. In the first 13 days of life the newborn received surfactant treatment combined with mechanical ventilation. Subsequently, bronchopulmonary dysplasia treatment was undertaken. She was fed exclusively parenterally until 15 days after birth, when, gradually, enteral nutrition was introduced through a nasogastric tube. After 16 weeks of hospitalisation the baby was discharged home where enteral nutrition through a nasogastric tube was continued. Until 5 MOA the baby was hospitalised on many occasions due to recurrent aspiration pneumonia and respiratory failure. At 5 MOA she was admitted to the Department of Anaesthesiology and Intensive Care for Children at the Pomeranian Medical University (PUM) Clinic in Szczecin due to worsening respiratory dysfunction, dyspnoea and a drop in O_2 saturation to 80%. Respiratory treatment was provided for 12 days. The therapy was accompanied by a number of other implemented treatments which included: invasive monitoring, intravenous fluid therapy, nutritional therapy, antibiotic treatment, nebulization, antiepileptic treatment as well as diuretic and protective treatment. Despite all the treatments and discontinued respiratory therapy, the baby was still fed through a nasogastric tube.

In the 13th day of the baby's stay at the Department of Anaesthesiology and Intensive Care for Children PUM neurological speech therapy was instituted. The relevant speech examination revealed impaired suck-swallow reflex. On top of that, the baby was also diagnosed with the absence of other essential reflexes such as the rooting reflex, gag reflex, bite reflex and oral reflexive movement. The muscles underneath the tongue were hypertonic, an uplift of the tip of the tongue to the hard palate during the abduction of the jaw and shifting the tongue back towards the soft palate and the pharyngeal wall. The girl received neurological speech therapy aimed at stimulating oral and rooting reflexes, reducing the hypertonia within the muscles located under the tongue, inducing the gag reflex as well as increasing mobility of the tongue and its proper frontal placement which was supposed to result in a better grip of the pacifier during nursing. 3 days after the commencement of the treatment, the gag reflex, the mandibular reflex, as well as occasional oral reflex were induced. After that, feeding with a regular teat was attempted with no apparent improvement in food intake. Then, in the fourth day of the treatment, the regular teat was replaced with one for babies with a cleft lip. It is characterized by a large external part, which proved helpful in upper lip stimulation and tight sealing of the mouth. A short internal section of the teat, through the licking reflex, contributes to the proper frontal placement of the tongue and facilitates food intake (Fig. 1).

The application of the new teat brought a considerable increase in the volume of a single food intake and better breathing (Fig. 2).

Subsequently, the baby was transferred to the Department of Paediatrics, Endocrinology, Diabetology, Metabolic Disorders and Cardiology of Developmental Age PUM for further neurological speech therapy and expansion of the diet. Two

Figure 1. Neurological speech therapy performer by means of a cleft lip teat.

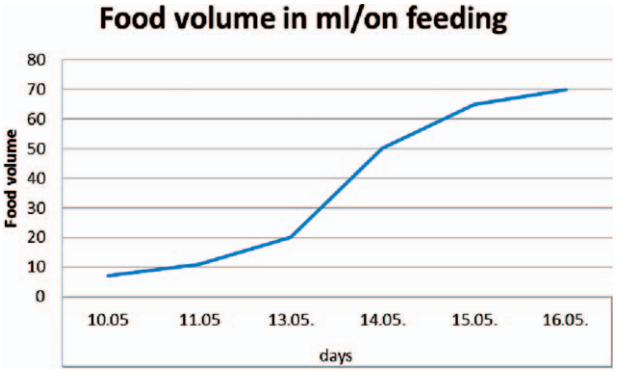

Figure 2. Distribution of daily food intake in milliliter in particular days of hospital stay.

weeks later the girl was discharged with satisfying therapeutic effect. On admission the baby weighed 3770 g and on discharge, 3 weeks later, the weight had increased to 4250 g.

3.2. Case presentation 2

Subject: 2.5-month-old baby boy, from pregnancy I, delivery I at a gestational age of 39 weeks, admitted to the Department of Paediatrics, Endocrinology, Diabetology, Metabolic Disorders and Cardiology of Developmental Age PUM with suspected infection of the upper airways. When interviewed the mother reported symptoms of reduced nasal patency of different severity that had persisted for about 1.5 months, intermittent hoarseness, sleep disturbances and apnea. The baby slept for very short periods, then woke up and cried. Having been breastfed for the first month the boy showed slow weight gain was, so the feeding was continued with Nutricia baby food Bebilon 1. Symptomatic treatment was carried out with Nasivin, Neomycin, Cebion, Aromactiv and 0.9% NaCl to no effect. While in hospital the baby presented irritability, difficulty breathing, reduced nasal patency with no discharge present, excessive drooling and occasional apnea. Psychomotor development corresponded with his age—he had good appetite and proper weight gain (85 centile). No vomiting or spitting up was observed. Auscultation revealed "nasal whir" while the lymphatic nodes were unenlarged. Both respiration and circulation were within the normal range, the skin was of a pale rose colour, the anterior fontanelle was even and muscle tone normal. The pharynx and tonsils were not inflamed, the heart rate regular and the vesicular murmur over the lung fields were physiological. Neither abdominal pain nor tenderness was detected. The laboratory test results were within normal limits (WNL), submucous cleft palate was ruled out and the otoscopic examination did not reveal any abnormalities. Still, a whirring noise could be heard in the nasal cavities despite the nostrils being unobstructed. A magnetic resonance imaging (MRI) examination of the nose and nosopharynx was performed. The ventricular system was symmetric, the ventricles were enlarged to 7 mm and the frontal horns to 4 mm. The nasal canals were properly developed and patent. However, the examination revealed a backward shift of the tongue towards the soft palate and the pharyngeal wall during the abduction of the jaw, lifting the tip of the tongue to the hard palate, overexpression of the jaw reflex and diminished expression of the oral reflex movements (Fig. 3).

The baby was administered a day long therapeutic speech massage aimed at lowering tone in the muscles below the tongue, drawing the tongue forward to its frontal position, reducing tension within the temporomandibular joint. A cleft lip teat was used for feeding (Fig. 4).

The following day mother noticed a significant improvement in breathing. The baby slept continuously for 5 hours at night. The "nasal cavity whirl" was also of reduced intensity. After 3 days' hospital stay the baby was discharged with recommendations for the use of a cleft lip bottle teat for another week and regular checkup visits. A week later the nasal sounds subsided, the baby's sleep was sound and breathing regular, undisturbed by apnea (Fig. 5).

4. Discussion

The use of teats for nutritive and a pacifier for non-nutritive sucking should follow detailed recommendations based on such criteria as: age (prematurely born infants), nutritional practices (contraindications to breastfeeding), presence of deficits (e.g., neurological deficit) or the baby's health. A bottle teat may prove invaluable in a situation when the baby is separated from their mother or when feeding requires therapeutic support.[16] There are still few scientific publications which would discuss the role of teats in infants afflicted with neurological dysfunctions, respiratory failure or insufficiency and sleep disorders.

Our study demonstrates that a proper application of stimulation with a cleft lip teat in infants suffering from respiratory and sleep disorders can bring improvement in performance of the respiratory system and the quality of sleep.

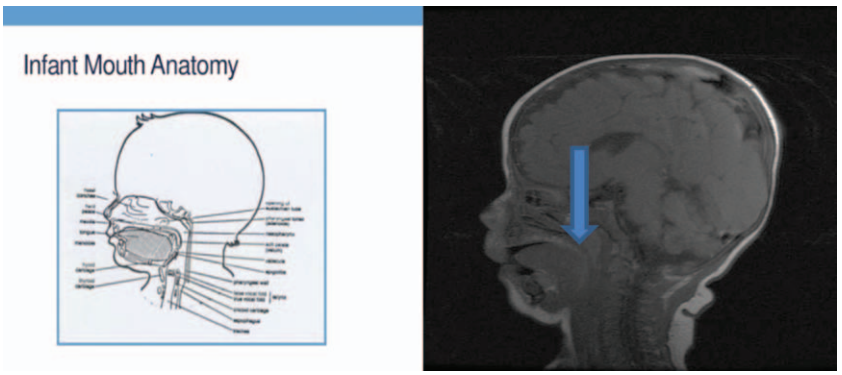

Figure 3. (A) Depicts a proper position of the oral cavity in infants;[15] (B) Shows an MRI cerebral image of 2.5-month-old baby with the tongue shifted back (arrow).

Figure 4. A cleft lip bottle teat.

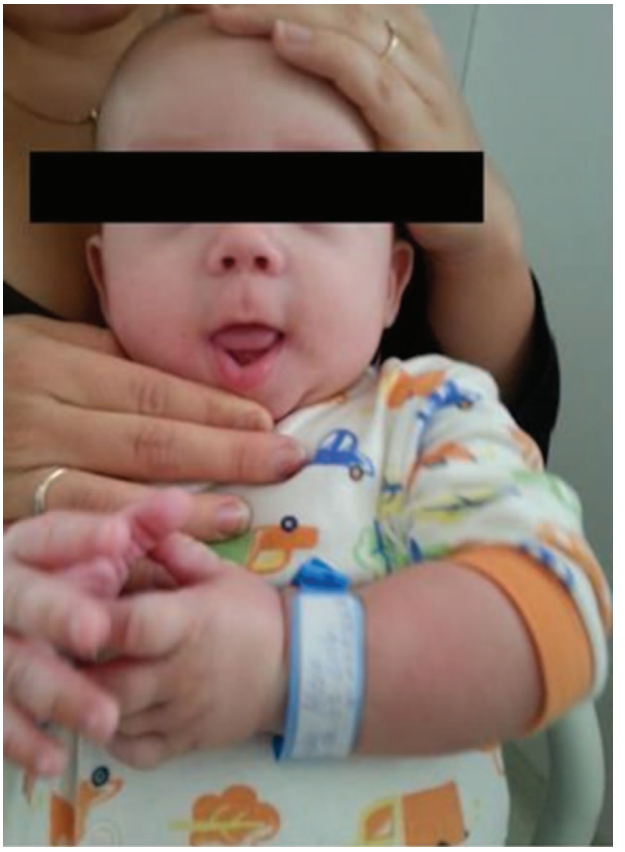

Figure 5. The baby's oral cavity after a week therapy with a cleft lip bottle teat.

These can be achieved through lowering the tone of the muscles under the tongue and increasing the tone of the orbicularis oris muscle.

Effective food intake in newborns and young babies is determined by proper oral stimulation aimed at activating relevant reflexes such as: the cough reflex, the gag reflex, the transversal tongue reflex/protrusion, the rooting reflex as well as the palmomental reflex. However, the key organ responsible for the quality of food intake and improving the reflexes of sucking, gagging and swallowing is the tongue, whose movements and actions are determined by a number of relevant muscles including the hyoglossus and genioglossus. It has been demonstrated that the hyoglossus plays a huge role in respiration,[17] as it retracts the protruded tongue and pulls it towards the floor of the mouth.

The genioglossus, one of the upper airways (UA) dilating muscles, clearly plays an important role in physiological maintenance of UA patency and pathophysiology of sleep-disordered breathing conditions, including obstructive sleep apnea (OSA).[18]

Research has also showed that the negative intrapharyngeal pressure is the main stimulus activating the pharyngeal muscles when one is waking up.[19] It is well known that the application of negative pressure to the pharyngeal airway in animals and in humans leads to a substantial increase in the activity of the genioglossus as well as other upper airway muscles.[19] Studies have recently shown that peak phasic genioglossus activity correlates closely with the peak negative epiglottic pressure generated during inspiratory resistive loading in healthy individuals.[20] It has also been suggested that during wakefulness genioglossal muscle activation is directly proportional to intra-pharyngeal pressure, probably secondary to activation of a local "negative-pressure reflex."[21]

The influence of sleep on the activation of the hyoglossus is very complex. During sleep healthy individuals exhibit a physiological reduction in the activity of the genioglossus muscle.[21] This finding has a number of implications. First, it suggests that during nonrapid eye movement (NREM) sleep, when the negative pressure reflex is substantially reduced, the genioglossus is minimally able to respond to increasing pharyngeal pressure and pharyngeal resistance. Thus, if the upper airway is dependent upon this reflex to maintain patency then collapse to some extent must ensue (as occurs in the apnoea patient).[22,23]

The babies participating in our studies demonstrated an increased tension of the genioglossus, which might explain why the children were aroused at night and could not go into a deep sleep (case 2). Moreover, higher tension in those muscles forced the posterior position of the tongue. These factors are most probably to blame for the disturbed sleep and sleep apnoea that affected the babies. Also, the baby boy did not have the adequate amount of sleep as he kept waking up during night time (case 2),

whereas the girl, due to the neurological deficits within the oral cavity, still required oxygen therapy (case 1). It was imposable to make an objective assessment of her sleep because of the pharmaceutical treatment she was receiving. The respiratory disorders that both infants presented could be the result of a lowered muscle tone in the anterior part of the oral cavity and consequently a reduced tone of the genioglossus. We believe that the shape of a specialist cleft lip teat, its wide base ensuring a good seal and a short top, stimulated the genioglossus, whose posterior horizontal fibres activated proper movement of the root of the tongue forward and up and the remaining fibres pulled the tongue down, off the palate, and pressed it to the bottom of the oral cavity. Lowering the dorsum of the tongue the horizontal fibres brought the tongue to the front. An argument supporting our observation is the fact that the tone of both muscles guarantees a proper position of the tongue. If the tone is reduced, then, at the horizontal position of the head (during sleep or lying), the tongue will drop backwards under its own weight, which most likely occurred in the infants we studied. A specialist cleft lip teat may prove to be a therapeutic tool accompanying neurological speech therapy in infants.

We are well aware a photograph of the tongue taken prior to the cleft lip teat therapy would have significantly contributed to reliability and validity of the study. Commencing the treatment we did not expect to receive such rapid progress and improvement within the oral cavity. We considered the application of a cleft lip teat to treat children with retracted tongues as a clinical trial and did not think of any photographic documentation at that time. The idea of a follow-up MRI procedure for the evaluation of the current tongue position must be ruled out due to the high risk the examination entails since it is carried out under general anesthesia to prevent movement artifacts. Recent research points to an increased risk of neuro-developmental disorders induced by applying anesthesia to babies.[24] Having this in mind we had no choice but to verify the effects of the therapy on the basis of the clinical picture of the patients. The positive effect of the therapy on the children's breathing and eating was not only observed by us but by the parents as well.

5. Conclusions

A specialist bottle teat designed for babies with cleft lips can constitute an effective tool in the therapy of nonspecific respiratory disorders resulting from improper position of the tongue and other articulatory organs.

Author contributions

Conceptualization: Wioletta Pawlukowska.
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Supervision: Iwona Rotter.
Writing – original draft: Wioletta Pawlukowska.
Writing – review & editing: Maria Giżewska.

References

[1] Ponti M. Recommendations for the use of pacifiers. Paediatr Child Health 2013;8:515–28.
[2] Bertoncelli N, Cuomo G, Cattani S, et al. Oral feeding competences of healthy preterm infants: a review. Int J Pediatrics 2012;2012:896257.
[3] Niemelä M, Pihakari O, Pokka T, et al. Pacifier as a risk factor for acute otitis media: a randomized, controlled trial of parental counselling. Pediatrics 2000;106:483–8.
[4] Als H, McAnulty GB. The newborn individualized developmental care and assessment program (NIDCAP) with kangaroo mother care (KMC): comprehensive care for preterm infants. Curr Womens Health Rev 2011;7:288–301.
[5] Thomson G, Bilson A, Dykes F. Implementing the WHO/UNICEFF baby friendly Initiative in the community:a "hearts ans minds" approach. Midwifery 2012;28:258–64.
[6] Sexton S, Natale R. Risks and benefits of pacifiers. Am Fam Physician 2009;79:681–5.
[7] Kramer MS, Barr RG, Dagenais S, et al. Pacifier use, early weaning, and cry/fuss behavior: a randomized controlled trial. JAMA 2001;286:322–6.
[8] Neiva FCB, Leone CE, Leone C, et al. Non-nutritive sucking evaluation in preterm newborns and the start of oral feeding: a multicenter study. Clinics 2014;69:393–7.
[9] Foster JP, Psaila K, Patterson T. Non- nutritive sucking for increasing physiologic stability and nutrition in preterm infants. Cochrane Database Syst Rev 2016;4:4–56.
[10] Nyqvist KH, Häggkvist AP, Hansen M, et al. Expansion of the baby friendly hospital initiative ten steps to successful breastfeeding into neonatal intensive care: expert group recommendations. J Hum Lact 2013;29:300–9.
[11] Jenik AG, Vain N. The pacifier debate. Early Hum Dev 2009;85:S89–91.
[12] Prade LA, de Paula BG, Weinmann ARM. The influence of behavioral state on sucking patterns in preterm infants. Audiol Commun Res 2014;19:230–5.
[13] Arvedson JC, Swallowing and feeding in infants and young chilren. GI Motility online; Available from 16 May 2006.
[14] Hunt CE. Infant sleeping position and SIDS risk. Pediatrics 1994; 94:105–7.
[15] Logemann JA. Evaluation and treatment of swallowing disorders. 2nd ed. Pro-Ed; Austin Texas: 1998.
[16] Lubbecorresponding W, Ham-Baloyi W. When is the use of pacifiers justifiable in the baby-friendly hospital initiative context? A clinician's guide. BMC Pregnancy Childbirth 2017;17:130Published online 2017 Apr 27.
[17] Stone M, Davis EP, Douglas AS, et al. Modeling the motion of the internal tongue from tagged cine-MR images. J Acoust Soc Am 2001;109:2974–82.
[18] Ono T. Tongue and upper airway function in subjects with and without obstructive sleep apnea. Jpn Dental SCi Rev 2012;48:71–80.
[19] Mathew OP, Abu-Osba YK, Thach BT. Genioglossus muscle response to upper airway pressure changes: afferent pathways. J Appl Physiol 1982;52:445–50.
[20] Horner R, Innes J, Morrell M, et al. The effect of sleep on reflex genioglossus muscle activation by stimuli of negative airway pressure in humans. J Physiol 1994;476:141–51.
[21] Malhotra A, Pillar G, Fogel R, et al. Genioglossal but not palatal muscle activity relates closely to pharyngeal pressure. Am J Respir Crit Care Med 2000;162:1058–62.
[22] Fogel RB, Malhotra A, Pillar G, et al. Genioglossal activation in patients with obstructive sleep apnea versus control subjects. Mechanisms of muscle control. Am J Respir Crit Care Med 2001;164:2025–30.
[23] Worsnop C, Kay A, Kim Y, et al. Effect of age on sleep onset-related changes in respiratory pump and upper airway muscle function. J Appl Physiol 2000;88:1831–9.
[24] Bartkowska-Śniatkowska A, Rosada-Kurasińska J, Zielińska M, et al. Do we really know the pharmacodynamics of anaesthetics used in newborns, infants and children? A review of the experimental and clinical data on neurodegeneration. Anaesthesiol Intensive Ther 2014;46:101–8.