CASE REPORT

THE PATIENT
6-day-old neonate

SIGNS & SYMPTOMS
– Failure to thrive
– Trouble feeding
– Intermittent retractions with inspiratory stridor

Benjamin P. Hansen, MD; David C. Fiore, MD; Kristine E. Galek, PhD
Renown Medical Group, Reno, Nev (Dr. Hansen); University of Nevada, Reno School of Medicine (Drs. Fiore and Galek)

Bhansen7000@gmail.com

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THE CASE
A primiparous mother gave birth to a girl at 38 and 4/7 weeks via uncomplicated vaginal delivery. Prenatal labs were normal. Neonatal physical examination was normal and the child's birth weight was in the 33rd percentile. APGAR scores were 8 and 9. The neonate was afebrile during hospitalization, with a heart rate of 120 to 150 beats/min and a respiratory rate of 30 to 48 breaths/min. Her preductal and postductal oxygen saturations were 100% and 98%, respectively. She was discharged on Day 2 of life, having lost only 3% of her birth weight.

The patient was seen in clinic on Day 6 of life for a well-child exam and was in the 17th percentile for weight. At another visit for a well-child exam on Day 14 of life, she had not fully regained her birth weight. At both visits, the mother reported no issues with breastfeeding and said she was supplementing with formula. The patient was seen again for follow-up on Days 16 and 21 of life and demonstrated no weight gain despite close follow-up with the Special Supplemental Nutrition Program for Women, Infants, and Children (WIC), which determined the newborn had some breastfeeding issues but seemed to be consuming adequate calories. However, WIC assessments revealed that during feeding, the child was expending too many calories and had nasal congestion. The patient was admitted to the hospital on Day 21 of life with a diagnosis of failure to thrive (FTT), at which point she was in the 12th percentile for weight.

THE DIAGNOSIS
Shortly after the infant was admitted, she showed signs of respiratory distress. On physical examination, the on-call resident noted intermittent retractions with inspiratory stridor, and the patient demonstrated intermittent severe oxygen desaturations into the 70s. She also was sucking her pacifier furiously, which appeared to provide some relief from the respiratory distress. The child's parents noted that she had demonstrated intermittent periods of respiratory distress since shortly after birth that seemed to be increasing in frequency.

Upon careful examination, the on-call resident identified a cystic lesion at the base of the child's tongue. The otolaryngologist on call was brought in for an urgent consultation but was unable to visualize the lesion on physical examination and did not recommend further intervention at that time. The patient continued to demonstrate respiratory distress with hypoxia and was transferred to the pediatric intensive care unit for close monitoring.

The next morning a second otolaryngology consultation was requested. A computed tomography scan of the neck demonstrated a 1.5-cm cystic-appearing mass at the base of the tongue that was obstructing the patient's airway. Direct flexible bronchoscopy confirmed the radiographic findings. The patient underwent immediate surgical resection of the lesion using a laser. A clear and milky gray cystic fluid exuded from the cyst when the lesion was pierced. The otolaryngologist visualized a widely patent airway following excision of the lesion (**FIGURE**).

Pathology results revealed no evidence of malignancy. The final diagnosis was a simple base-of-tongue cyst.

DISCUSSION

Failure to thrive is common in neonates and occurs most often due to inadequate caloric intake; however, it also can be caused by systemic disease associated with inadequate gastrointestinal absorption or increased caloric expenditure, such as congenital heart disease, renal disease (eg, renal tubular acidosis), chronic pulmonary disease (eg, cystic fibrosis), laryngomalacia, malignancy, immunodeficiency, or thyroid disease.[1]

Respiratory distress in neonates also is common but tends to occur shortly after birth.[2] Conditions associated with respiratory distress in neonates include transient tachypnea of the newborn, respiratory distress syndrome, pneumothorax, persistent pulmonary hypertension of the newborn, pneumonia, and meconium aspiration syndrome.[2] Interestingly, there are additional reports in the literature of FTT and respiratory distress in neonates caused by obstructive pharyngeal lesions.[3-5]

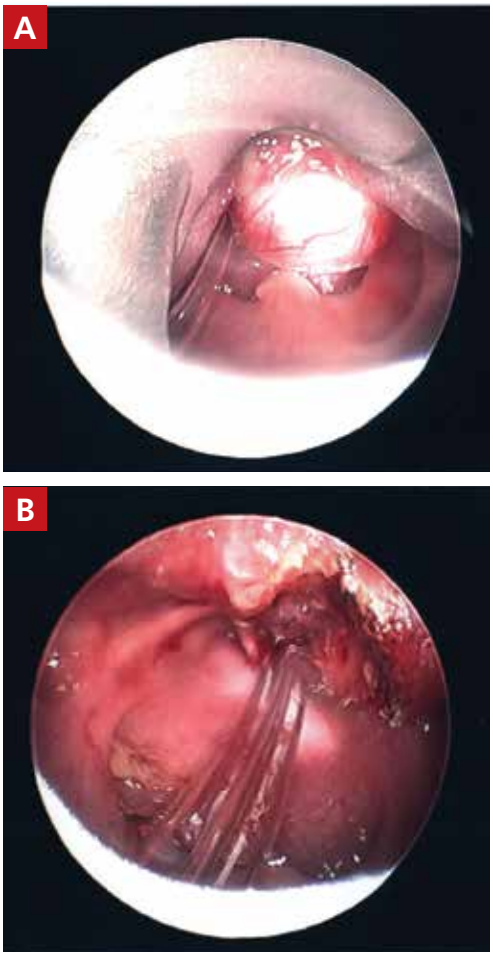
Base-of-tongue cysts are rare in infants. Fewer than 50 cases were reported prior to 2011, with many being described as asymptomatic nonpainful lesions.[6] Given the anatomic location of base-of-tongue cysts, the differential diagnosis should also include mucoceles, thyroglossal duct cysts, dermoid cysts, epidermoid cysts, vallecular cysts, hemangiomas, cystic hygromas, lymphangiomas, thyroid remnant cysts, teratomas, and hamartomas.[4,7,8] When tongue cysts are initially discovered, inspiratory stridor, FTT, swallowing deficits, oxygen desaturation, respiratory failure, and/or acute life-threatening events have been reported.[6,9,10]

One important clinical observation made in our case was the use of an external apparatus to relieve the neonate's respiratory distress. During physical examination, the on-call resident noted the patient was furiously sucking her pacifier, which seemed to reduce the respiratory difficulty and desaturations. It is known that non-nutritive sucking (NNS) can provide provisions for stress relief, improve oxygenation, and provide proprioceptive positioning of key anatomical structures within the oral cavity.[11] Without the use of an external apparatus like a pacifier during restful states, neonates may develop vacuum-glossoptosis syndrome, in which the dorsum of the tongue and the soft palate adhere to the posterior pharyngeal wall and obstruct the airway.[12] Our patient may have used the pacifier as an NNS task to move the tongue forward and break the glossoptosis-pharyngeal seal by sucking hard and fast during periods of respiratory distress, which reduced the potential for a vacuum-glossoptosis phenomenon that was likely created by the cyst during restful states.

Our patient was seen in clinic for follow-up after surgery on Day 35 of life. She

FIGURE

Surgical resection of the base-of-tongue cyst



IMAGES COURTESY OF: COURTNEY GARRETT, MD

Intraoperative photographs demonstrating the obstructive base-of-tongue cyst just prior to surgical resection (**A**) and the area immediately after resection (**B**).

Mechanical obstruction should be considered in neonates with failure to thrive and respiratory distress.

was thriving and her weight was in the 24th percentile. She was seen again on Day 67 of life for a well-child exam and was in the 43rd percentile for weight.

THE TAKEAWAY

There is a sizeable list of possible diagnoses to consider when a neonate presents with FTT and respiratory distress. It is important to consider mechanical obstruction as a possible diagnosis and one which, if identified early, may be lifesaving. Our case demonstrates a proposed mechanism by which a mechanical obstruction such as a base-of-tongue cyst can cause the vacuum-glossoptosis syndrome; it also highlights NNS as a potential means of overcoming this phenomenon. JFP

Non-nutritive sucking with a pacifier may relieve airway obstruction caused by base-of-tongue cysts.

CORRESPONDENCE
Benjamin P. Hansen, MD, Renown Medical Group, 4796 Caughlin Pkwy, Ste 108, Reno, NV 89519; Bhansen7000@gmail.com

References

1. Larson-Nath C, Biank VF. Clinical review of failure to thrive in pediatric patients. *Pediatr Ann*. 2016;45:e46-e49.
2. Edwards MO, Kotecha SJ, Kotecha S. Respiratory distress of the term newborn infant. *Paediatr Respir Rev*. 2013;14:29-37.
3. Brennan T, Rastatter JC. Multilevel airway obstruction including rare tongue base mass presenting as severe croup in an infant. *Int J Pediatr Otorhinolaryngol*. 2013;77:128-129.
4. Gutiérrez JP, Berkowitz RG, Robertson CF. Vallecular cysts in newborns and young infants. *Pediatr Pulmonol*. 1999;27:282-285.
5. Wong KS, Huang YH, Wu CT. A vanishing tongue-base cyst. *Turk J Pediatr*. 2007;49:451-452.
6. Aubin A, Lescanne E, Pondaven S, et al. Stridor and lingual thyroglossal duct cyst in a newborn. *Eur Ann Otorhinolaryngol Head Neck Dis*. 2011;128:321-323.
7. Hur JH, Byun JS, Kim JK, et al. Mucocele in the base of the tongue mimicking a thyroglossal duct cyst: a very rare location. *Iran J Radiol*. 2016;13:4-7.
8. Tárrega ER, Rojas SF, Portero RG, et al. Prenatal ultrasound diagnosis of a cyst of the oral cavity: an unusual case of thyroglossal duct cyst located on the tongue base [published online January 21, 2016]. 2016;2016:7816306.
9. Parelkar SV, Patel JL, Sanghvi BV, et al. An unusual presentation of vallecular cyst with near fatal respiratory distress and management using conventional laparoscopic instruments. *J Surg Tech Case Rep*. 2012;4:118-120.
10. Sands NB, Anand SM, Manoukian JJ. Series of congenital vallecular cysts: a rare yet potentially fatal cause of upper airway obstruction and failure to thrive in the newborn. *J Otolaryngol Head Neck Surg*. 2009;38:6-10.
11. Pinelli J, Symington A. Non-nutritive sucking for promoting physiologic stability and nutrition in preterm infants. *Cochrane Database Syst Rev 2005*. 2010;4:CD001071.
12. Cozzi F, Albani R, Cardi E. A common pathophysiology for sudden cot death and sleep apnoea. "the vacuum-glossoptosis syndrome." *Med Hypotheses*. 1979;5:329-338.