

Title:

Successful Long-Term Airway Stabilization with a Modified Pacifier in a Syndromic Infant

Authors:

Mehmet N. Cizmeci¹, M.D

Mehmet K. Kanburoglu¹, M.D

Selma Ziraman¹, R.N.P

Mustafa M. Tatli¹, M.D, Clinical Professor of Neonatology

Institution:

¹Division of Neonatology, Department of Pediatrics, Fatih University Medical School, Ankara

Corresponding Author:

Mehmet Nevzat Cizmeci, M.D

Department of Pediatrics, Division of Neonatology

Fatih University Medical School, 06510, Emek, Ankara/Turkey

Tel: +90 505 6576539

e-mail: nevzatcizmeci@gmail.com

Conflicts of Interest

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Abstract

Airway management is one of the key aspects of neonatal anesthesia and an oropharyngeal airway (OPA) is used in daily intensive care practice to relieve upper airway obstruction. A myriad of congenital malformations have features that might cause a difficult airway management and cleft palate is among these conditions. Herein, we describe a syndromic infant with alobar holoprosencephaly whose long-term airway patency was maintained with a modified infant pacifier. We would like to share our experience and contribute to the literature with the introduction of a readily available and easily-inserted apparatus.

Keywords: Airway stabilization, neonate, oropharyngeal airway, syndromic infant.

Introduction

Airway management is one of the key aspects of neonatal anesthesia and an oropharyngeal airway (OPA) is used in daily intensive care practice to relieve upper airway obstruction. An OPA is a medical apparatus used to maintain a patent airway by preventing the tongue from obstructing the aperture of the upper airway structures.^{1,2}

A myriad of congenital malformations have features that might cause a difficult airway management due to either inability to optimally position the head or maldevelopment, which makes it difficult to obtain a good view of the upper airway structures. Cleft palate is among these conditions.^{3,4} Herein, we describe a syndromic infant with alobar holoprosencephaly whose long-term airway patency was maintained with a modified infant pacifier.

Case Summary

A male infant was born at 40 weeks' gestation to a 24-year-old gravida 1, para 1 healthy mother by cesarean section. The infant was detected to have holoprosencephaly and significant hydrocephalus in the first trimester. There was no consanguinity and the pregnancy history was unremarkable. The infant was admitted to the neonatal intensive care unit for adequate life support upon delivery.

There was significant microcephaly together with dysmorphic facial features including a blind-ended single nostril, ocular hypotelorism and complete cleft palate, all of which were consistent with cebocephaly. The rest of the physical examination was unremarkable. Magnetic resonance imaging (MRI) of the cranium was obtained to guide the modality of airway stabilization, which revealed an osseous septum in the posterior nasopharynx together with intracranial abnormalities consistent with alobar holoprosencephaly. Further imaging studies including echocardiography and abdominal ultrasonography were not remarkable;

however the electroencephalography (EEG) revealed diffuse repetitive electrographic seizures, which responded well to phenobarbital therapy.

Consultation was made with the otolaryngology clinic for the selection of the modality of the airway stabilization and a possible surgical procedure; however, the otolaryngology council rejected to perform a surgical operation owing to the risks associated with a procedure and lack of life expectancy related with the severe malformation.

Since the infant was an obligate mouth-breather and desaturated rapidly without any intervention, an OPA was first used to obtain the airway patency. Then, it was replaced by an infant pacifier that was one-size larger than appropriate for the infant's age. This pacifier had a hand-made hollow in the middle of the distal tip of the bulbous region. Air passage was obtained by removing the handle from the proximal tip of face plate (Figure 1a, 1b). The larger size of the pacifier provided an extension further into the anatomical airway than does a typical infant pacifier. The non-resilient polyethylene material ensured the airway patency and it did not collapse under force applied by the infant's bite. The modified apparatus was easily tolerated by the unsedated infant while having a soothing effect (Figure 1c). The patient's oxygen saturation stabilized at > 95% in room air without any further intervention. Since the infant was an obligate mouth breather, a percutaneous gastrostomy tube was placed for nutrition and he was discharged on the 21st postnatal day to be followed up by a multidisciplinary outpatient clinic.

Discussion

Airway devices play an important role in the acute management of patients and their use is especially recommended during difficult airway management in the operating room and in the pre-hospital setting.⁵ Today, novel airway devices are becoming widely used in routine practice.⁴ While we are in need to update our knowledge on the novel technologies, we should also disseminate simple and practical approaches that raise the quality of life, especially of infants with chronic and lethal conditions. Although an OPA may be used for this purpose, it is generally preferred for short term airway management. Our aim of using a modified infant pacifier was to prepare an infant with a major neurologic disability for discharge from the hospital, since the infant had an osseous septum in the posterior nasopharynx and could not tolerate the removal of an OPA. The apparatus was well-tolerated by the infant and maintained a long term patency of the upper airways.

After discharge, the parents were able to maintain the use of the device at home and the infant was seen regularly in the outpatient clinic for the follow-up visits. Repeated attempts to remove the apparatus, resulted in prominent respiratory distress. The infant is still alive and he is 5 months old at present, suffering mainly from frequent upper respiratory tract infections which may be attributable to the artificial patency of the airways.

We believe that unconventional approaches might increase the quality of life of patients with severe conditions related with a short life expectancy. We would like to share our experience and contribute to the literature with the introduction of this readily available and easily-inserted apparatus.

Ethical Approval

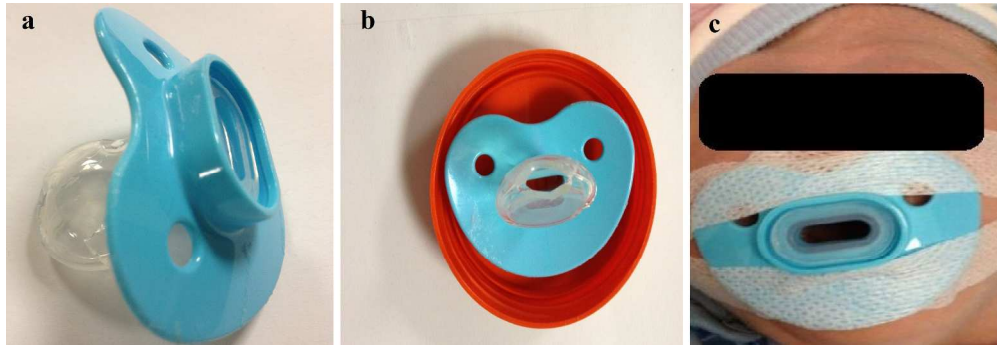
For this type of case report, no ethical approval is required in Turkey. Informed consent was obtained from the parents.

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Figure Legends

Figure 1: Lateral view of the pacifier with the handle removed (a). Proximal bulbous portion of the pacifier placed on a red surface to demonstrate the apertures of the airway conduit (b). The infant, while being adapted to the modified pacifier (c).



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