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Short Communication

Primary respiratory support in preterm infants with cleft lip and palate

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Abstract

Objective: Preterm infants with respiratory distress are routinely treated by application of nasal mask continuous positive airway pressure. In preterm infants with cleft lip and palate, nasal mask attachment is not feasible due to air leakage through the cleft defect. Here, we describe a modified continuous positive airway pressure application method that overcomes this problem.

Design and subjects: Observation study, university neonatal intensive care unit. The neonates ($n = 4$) were between 28 and 33 weeks' gestation and weighed 1160 to 1680 g at birth. Immediately after birth, infants with unilateral cleft lip and palate ($n = 3$) were respiratory stabilised by a Medijet^{TM} generator using a nasal tube. To minimise the pressure cap, hydrocolloid bandages were adhered over the total cleft defect. Immediately after orthodontic passive palatal plates insertion (within 27 hours of life), the nasal tube was removed and continuous positive airway pressure was applied through a nasal mask covering the complete nose of the infant.

Results: The system proved suitable for patients with unilateral cleft lip and palate for whom the generated nasal mask continuous positive airway pressure remained constant between 5 to 7 cm of water but failed in the patient with bilateral cleft lip and palate. None of the patients had to be ventilated due to respiratory failure, and all survived to discharge.

Conclusion: Preterm infants with unilateral, but not bilateral cleft lip and palate, can be successfully stabilised using the described nasal mask continuous positive airway pressure system, thereby avoiding primary intubation and its associated risk of complications.

Key words: Cleft Lip; Cleft Palate; Premature Infant; Continuous Positive Airway Pressure Ventilation

Introduction

Respiratory distress syndrome in newborn and preterm infants is widely treated with nasal continuous positive airway pressure (nCPAP), thereby avoiding the need for endotracheal intubation and mechanical ventilation.[1] Recently, McMahon and colleagues were the first authors who reported on a preterm infant with bilateral cleft lip and palate who was successfully given nCPAP as a method of respiratory stabilisation after initial mechanical ventilation.[2] We report on our experiences using a modified method of nCPAP application in four preterm infants with uni- and bilateral cleft lip and palate suffering from respiratory distress after being born.

Materials and methods

The neonates (Table I) were between 28 and 33 weeks' gestation, weighed between 1160 and 1680 g at birth, and were all inborns ($n = 4$). The antenatal period was uneventful in all pregnancies. Karyotyping showed a normal chromosome complement (fluorescent in situ hybridisation analysis) in all patients. Deliveries were performed via primary caesarean sections and mothers were given prenatal corticosteroids at least once (betamethasone, 2×8 mg, 24 hours prior to delivery). The use of our nasal continuous positive airway pressure system involves the application of the air stream with (a) a nasopharyngeal tube (Medijet 1020^{TM}, Medical Innovations, Puchheim, Germany) or (b) short prongs or masks of different sizes (Infant Flow^{TM} system, EME Ltd, Birmingham, UK) to the nose of the infant. Immediately after birth, infants with unilateral cleft lip and palate (patients one to three) were respiratory stabilised by a discontinuous system of nCPAP using a Medijet^{TM} generator connected to a shortened paediatric endotracheal tube (3 mm inner diameter, Vygon, Écouen, France) by a mononasal adapter (B + P Ltd, Neunkirchen-Seelscheid, Germany) placed into the normally formed contralateral orifice of the nose (Figure 1). The insertion depth of the tube was determined by the estimated distance between nose tip and ear to ensure that the position was not too deep (nasopharyngeal space). Note that thinner nasal tubes (2.5 mm inner diameter, Vygon, Écouen, France) were used for smaller babies (maximum weight 1250 g). The positive pressure was induced by a narrow stream of warmed humidified air/oxygen mixture generated by the Infant-Flow^{TM} driver. This gas is then inspired while expired gas is vented to the surrounding atmosphere. Flows ranging from 6 to 14 l/min allowed end expiratory pressure in the infant's upper airway of between 2 and 8 cm of water depending on the gas flow.[3] To minimise air leakage and loss in pressure caused by unilateral cleft lip and palate,

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TABLE I

PATIENT'S CHARACTERISTICS

Patient	1	2	3	4
Gestational age (wk), sex (m/f)	30 5/7, m	27 3/7, m	33 0/7, f	33 0/7, f
Birth weight (g)	1480	1160	1680	1440
Apgar (1,5,10 min)	8,9,9	3,7,8	8,9,9	8,9,9
Cleft side	Left	Right	Left	Both
Additional surfactant therapy (InSurE)	+	+	–	–
Complications	RDS	RDS, Infection	RDS	Wet lung
Time of insertion of orthodontic appliance (hours)	27	26	24	23
Total days of nCPAP therapy	3	19	2	2 h
Total days of oxygen demand	8	45	3	2 h
Positive end expiratory pressure	5	7	6	–
Additional intubation	–	+	–	–
IVH	none	none	none	none
Outcome	Survived	Survived	Survived	Survived

m = male; f = female; InSurE = intubation surfactant extubation; RDS = respiratory distress syndrome; IVH = intraventricular haemorrhage; nCPAPR = nasal continuous positive airway pressure

(a) hydrocolloid bandages (Varihesive™, ER Squibb & Sons, ConvaTec, Munich, Germany) were adhered over the total cleft defect and (b) straps were bound around the mandible and forehead to keep the mouth closed.

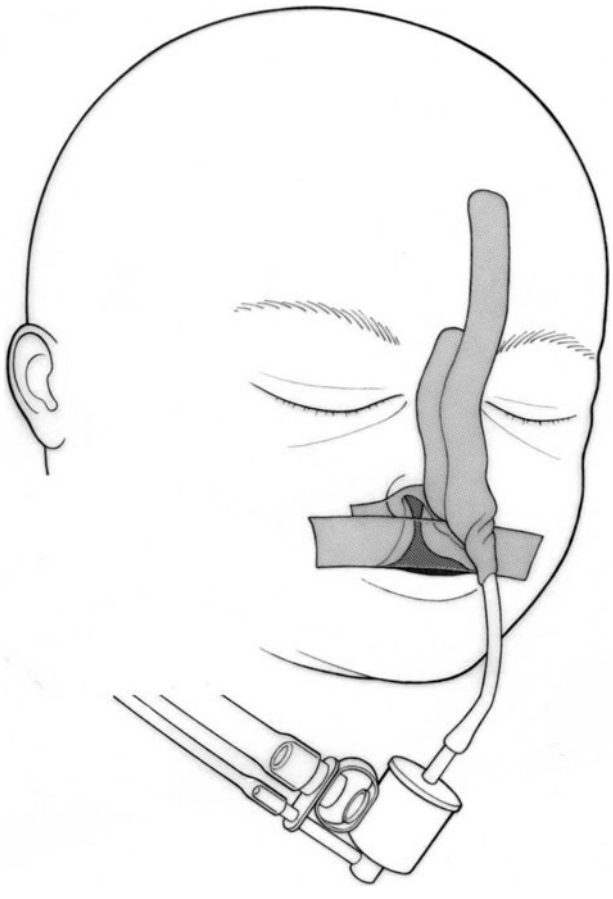

FIG. 1

Schematic drawing of the described nasal continuous positive airway pressure (nCPAP) system providing primary respiratory support for preterm infants with unilateral cleft lip and palate. This support is administered through a Medijet™ generator connected to a shortened paediatric endotracheal tube (3 mm inner diameter) placed into the normally formed contralateral nostril. To minimise air leakage and loss in pressure hydrocolloid bandages were adhered over the total cleft defect (interrupted lines).

In patient four, with a bilateral cleft lip and palate, no tube could be inserted and fixed into the nose. Therefore, to provide nCPAP, a silicone infant mask connected to the Infant-Flow™ driver was used as described by McMahon and colleagues.[2]

Patients one and two received a single endotracheal instillation of surfactant as primary respiratory support (Survanta™, Abbott Ltd, Wiesbaden, Germany; 100 mg/kg bw) within the first 30 minutes (via short-term intubation and immediate extubation) because of developing respiratory failure.[4]

Within the first 27 hours of life, orthodontic passive palatal plates were adjusted in all patients under spontaneous breathing supported by nasal tube CPAP as described above. Patient two had to be secondarily intubated and ventilated from the 21st to 68th hour of life due to superimposed infection. Immediately after insertion of the passive palatal plate, nasal tubes were removed for patients one and three, and in patient two after extubation, and a nCPAP-generator was applied with a mask covering the complete nose of the infant (Figure 2). Without any further complications nCPAP could be administered over several days (2–19 days, Table I). The generated positive airway pressure remained constant between 5 and 7 cm of water with an accommodated flow of 10 to 13 l/min. Oxygen demand varied between 25 to 30 per cent to reach at least a peripheral saturation of more than 92 per cent.

All four patients survived to discharge from the hospital and are currently undergoing serial clinical follow up. None of our four patients had evidence of bronchopulmonary dysplasia or retinopathy of prematurity. Ultrasonic investigations of the brain were normal. All patients had no evidence of additional nasal deformities or damage due to nasal mask or palate plate. Brainstem auditory evoked response testing did not identify hearing disorders at the age of two months in all patients.

Discussion

The challenge is how to secure effective air supply to patients, such as preterm infants, with cleft lip and palate requiring primary respiratory support. Mechanical ventilation is one possible option; however, it may be associated with lung barotrauma and risk of ventilator-induced lung injury. Another option is the application of nasal continuous positive airway pressure for the management of respiratory failure. However, common continuous positive airway pressure application systems cannot be used because the

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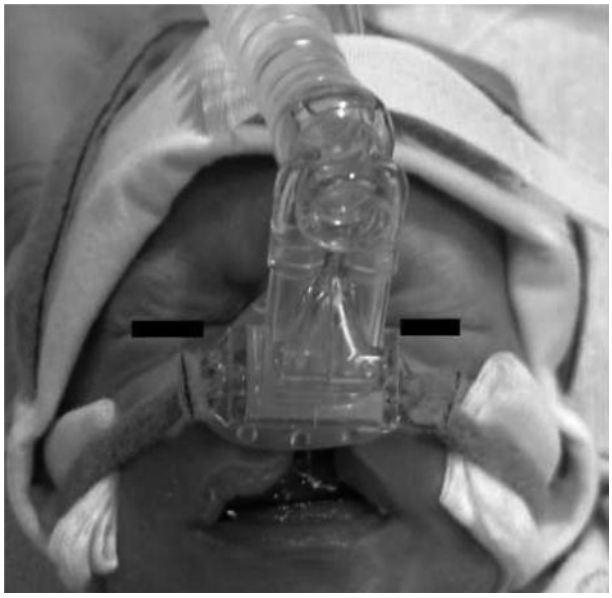

FIG. 2
Patient three at third day of life with Infant Flow™ and inserted palate plate.

system should be tightly closed and the generated positive airway pressure should not leak out through the open mouth or nose of the patients.

Therefore, to better treat these patients with cleft lip and palate, we modified the nCPAP application system. As described, the inserted tube should be as large as possible and shortened to minimise the tube resistance that limits the effectiveness of CPAP. Pressures of 5 to 7 cm of water are recommended. To minimise necrosis of the nasal mucosa and conchae due to the mechanical pressure of the inserted nasal tube, early removal followed by the use of a nasal mask is desirable.[5] However, before a mask can be used, the passive palatal plate must be inserted as soon as possible and has to be adjusted to avoid air leakage through the cleft and dislocated hypoplastic maxillary segments which could otherwise result in alveolar collapse. The orthodontic appliance is a plate of hard acrylic resin and is designed as described by Hotz and Gnoinski.[6]

The airway is smallest in patients with unilateral cleft lip and palate and is largest in those with bilateral clefts.[7] Hence, in such cases, it is not only imperative that the palate plate be accurately adjusted to take into consideration the cleft, but it must also accommodate the smaller airways in preterm patients as well. However, despite the much higher prevalence of oral breathing in cleft infants compared to non-cleft, we did not observe any complications in application of nasal breathing assistance.[8]

We did not apply nCPAP to patient four with bilateral cleft lip and palate because the nasal tube could not be fixed. It may be advisable to intubate this special group of patients upon signs of beginning respiratory distress or perform mask CPAP as described by McMahon and colleagues.[2] After this, extubation and nCPAP can be performed in the same manner as described above. Therefore, the application system seems best suited for treatment of unilateral cleft defects.

Finally, the described nCPAP application system caused no severe complications, such as pneumothoraces, pneumopericardium, intestinal complications, and nasal deformities, secondary to the nares-occluding prongs, and can be used for primary respiratory stabilisation and support in the delivery room just after the child is born.

Conclusion

Preterm infants with unilateral cleft lip and palate suffering from respiratory distress can be stabilised by using a modified nasal application of the continuous positive airway pressure (CPAP) system. This system consists of applying nasopharyngeal CPAP with a tube, early adjustment of the passive palatal plate, and then nasal continuous positive airway pressure (nCPAP) applied with a mask. The described nCPAP application system circumvents the need for primary intubation. Finally, this application method is more comfortable for the patients and is not associated with increased risk of disease or other negative complications.

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