

Indirizzo dell'autore: DR. EDOARDO BERNKOPF
VICENZA 36100- Via Garofolano, 1 - Tel. 0444/545509
PARMA 43100- Via Petrarca, 3 - Tel. e Fax 0521/236426
ROMA 00100- Via Massaciucoli, 19 Tel. 06/86213428
E-mail: edber@studiober.com
Sito internet: <http://www.studiober.com>

AMERICAN JOURNAL OF RESPIRATORY AND CRITICAL CARE MEDICINE VOL 166 2002

Correspondence

1. Li KK, Riley RW, Guilleminault C. An unreported risk in the use of home nasal continuous positive airway pressure and home nasal ventilation in children. *Chest* 2000;117:916-918.
2. Villa MP, Dotta A, Castello D, Piro S, Pagani J, Palamides S, Ronchetti R. Bi-level positive airway pressure (BiPAP) ventilation in an infant with central hypoventilation syndrome. *Pediatr Pulmonol* 1997;24:66-69.
3. Delaire J. Maxillary development revisited: relevance to the orthopaedic treatment of Class III malocclusions. *Eur J Orthod* 1997;19:289-311.
4. Kilicoglu H, Kirlic Y. Profile changes in patients with class III malocclusions after Delaire mask therapy. *Am J Orthod Dentofacial Orthop* 1998;113:453-462.
5. Takada K, Petdachai S, Sakuda M. Changes in dentofacial morphology in skeletal Class III children treated by a modified maxillary protraction headgear and a chin cup: a longitudinal cephalometric appraisal. *Eur J Orthod* 1993;15:221-222.

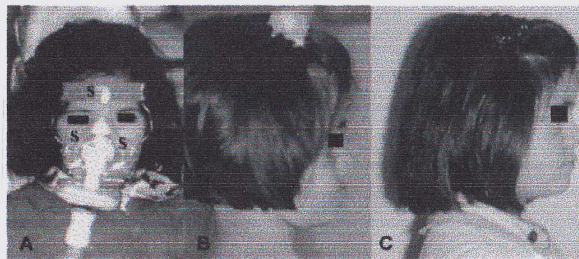


Figure 1. (A) photograph showing Delaire mask mounted on the nasal BiPAP mask with screws (S). (B) Photograph showing a profile of the patient before treatment with the modified Delaire mask. Note the deformed mid-face. (C) Photograph taken after treatment showing the facial structure returned to normal.



PRIMA DEL
TRATTAMENTO

DOPO IL
TRATTAMENTO

MID-FACE HYPOPLASIA AFTER LONG-TERM NASAL VENTILATION

To the Editor:

Continuous positive airway pressure (CPAP) and bilevel positive airway pressure (BiPAP) ventilation provide life support in most chronic respiratory diseases. In many cases, treatment starts early in life. In young children, noninvasive mechanical ventilation is increasingly delivered through a nasal facemask. Because at the age of 4 years 60% of the adult face has developed, early long-term treatment using a nasal mask carries a high risk of facial growth impairment. In a recent case report, Li and colleagues (1) noted that the force exerted by the mask during long-term use of nasal BiPAP could retard facial development. In this letter, we describe the first case treated successfully with a corrective device. As previously reported (2), the girl, who is now 7 years old, began receiving ventilation therapy with nasal BiPAP for central hypoventilation syndrome (CHS) at the age of 9 months. The child's mother had refused permission for tracheotomy. CHS is defined as a failure in the automatic control of breathing that is present from birth, is characterized by hypoventilation that worsens during sleep, and is of unknown etiology. The girl has now developed normally both physically and psychologically, and all functional data suggest normal development. At the latest assessment, at the age of 7 years, the child had visible maxillary hypoplasia, not recognizable at previous assessments. After a diagnosis of mid-face hypoplasia and Angle Class III malocclusion based on a cephalometric evaluation, the orthodontist recommended starting orthopedic therapy. Presumably, during prolonged nasal BiPAP therapy, pressure of the headgear

and face mask unit, especially against the malleable nasal, zygomatic and maxillary area, had interfered with mid-face development. No members of the child's family had orthodontic anomalies.

To solve the problem of continuing nasal BiPAP without compromising normal facial development we mounted a common Delaire mask (Figure 1A), a standard orthodontic device in the treatment of Angle Class III malocclusion and severe maxillary hypoplasia (3, 4), onto the nasal BiPAP mask. The Delaire mask was mounted on the nasal BiPAP mask with screws (Figure 1A). The custom-designed screw device on the Delaire mask could be tightened or loosened so that the orthodontist could, when necessary, alter the distance between the two masks and minimize pressure of the mask on the face. It also allowed the mask to be fitted tightly over the patient's face to prevent air leaks.

The modified Delaire-BiPAP mask, applied to the patient's face and fixed to the upper dental arch with elastic bands, exerted suitable corrective orthopedic traction on the zygomatic-maxillary area, while the face-gear exerted a counteracting force on the chin (4) and forehead. The modified Delaire mask was applied at night and during the afternoon nap, for about 10 hours, while the child was being ventilated with nasal BiPAP. Within 10 months, the maxillary hypoplasia and the mandibular protrusion had visibly diminished (Figures 1A and 1B) and the cephalometric index normalized (subspinal point to nasion to supramental point angle: before treatment = -2.7° , after treatment = -1.5° ; sella to nasion to subspinal point angle: before = 82.4° , after 83° ; sella to nasion to supramental point angle: before = 85.1° , after = 84.4°). This improvement agrees with published treatment times for children with Angle Class III malocclusion without nasal BiPAP treated with the Delaire mask (5). Because the early assessments disclosed no evidence of maxillary anomalies, the child did not initially undergo orthodontic follow-up. Therefore, we cannot say exactly when her mid-face hypoplasia began. The meaning of "long-term" ventilatory therapy is hard to define. In our case with a growing child, 6 years probably sufficed for the anomalies to develop, insofar as this period coincided with maximum skeletal growth. Apart from the duration of treatment, another cause of the ventilation-induced adverse effects in our patient could be the ventilatory pressures. The girl still wears the Delaire mask mounted onto the nasal ventilation mask. The combined appliance seems to achieve good ventilatory and orthodontic-orthopedic compensation. Our case again underlines the possible adverse effects of long-term mechanical ventilation on craniofacial development. To avoid these complications, growing children receiving long-term ventilation therapy via a nasal mask should undergo regular orthodontic surveillance.

MARIA PIA VILLA

JACOPO PAGANI

ROSA AMBROSIO

ROBERTO RONCHETTI

University of Rome, Sant'Andrea Hospital
Rome, Italy

EDOARDO BERNKOPF
Vicenza, Italy