Iatrogenic Nasal Vestibular Stenosis: A Rare Cause of Apnea

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Abstract
Nasal vestibular stenosis is a rare complication following supportive care in premature infants, causing airway and feeding difficulties in the obligate nasal breather. The imputation of the nasal continuous positive airway pressure (CPAP) in this type of complication has been increasingly described since this ventilatory mode provides an effective alternative and is routinely used at many neonatal resuscitation care units (NRCU). In the present study we describe a case of a premature neonate with severe apneas since the age of 21 days due to iatrogenic nasal synchia, following use of nasal CPAP for respiratory distress that was successfully repaired with endoscopic lysis and repeated dilatations.

Keywords: Neonate; Apnea; Nasal CPAPA; Synchia

Abbreviations: CPAP: Continuous Positive Airway Pressure; NRCU: Neonatal Resuscitation Care Units; NICU: Neonatal Intensive Care Unit.

Case Report
Z.H is a female premature neonate born at 31 weeks 1 day of gestation from a consanguineous marriage via spontaneous vaginal delivery with a breech presentation [1,2]. The mother does not have a medical history (25 years, gravida I, para I, Blood group O +). Pregnancy had been normal. Routine prenatal ultra-sonograms had been normal. At birth the neonate weighed 1200 g, and Apgar scores were 7 and 8 at one and five minutes. The newborn was in respiratory distress with perioral cyanosis and a silver man score of 5. Nasal catheters were passed without difficulty through both nasal passages. She was admitted to the neonatal intensive care unit (NICU), placed in a preheated incubator with cardiorespiratory monitoring. Birth examination at the unit showed perioral cyanosis, oxygen saturation at 80%, heart rate 110/min, no heart murmur, recolouring time<3s, present and symmetrical pulse. Chest x-ray showed a bilateral alveolar syndrome and poor chest expansion evoking a hyaline membrane disease at an advanced stage requiring supplemental oxygen administration with nasal CPAP and instillation of two doses of surfactant. She was first perfused via an umbilical venous catheter and then a transjugular catheter. She was put on probabilistic antibiotic therapy for suspected materno fetal infection nullified after investigations. Milk was introduced progressively on the 3rd day of life and then a transjugular catheter. She was put on ventilatory support for respiratory distress that was successfully repaired with endoscopic lysis and repeated dilatations.

Z.H was transferred to the breeding unit. On the 21st day of life she had begun to make iterative apneas especially during sleep and feeding. The explorations were negative (Infectious tests<0, serum calcium and glycemia were normal, Chest x-ray, transfontanellar ultrasound and cardiac ultrasound were normal also). A nasal airway obstruction had been evoked since the neonate had bilateral nasal vestibular retraction and it was impossible to introduce a nasal catheter through nasal passages. A facial mass scan was performed...
and it was normal. A nasal endoscopy was performed and showed nasal synechia obscuring 80% of the nasal airway bilaterally. The newborn received synecholysis and repeated dilatations. Currently 9 years passed, the patient remained asymptomatic, without recurrence of synechia.

**Discussion**

Neonates are obligatory nasal breathers so that’s why nasal obstruction results in respiratory distress, apnea, cyanosis, feeding and sleep difficulties and in long-term a failure to thrive. Such characteristic remains on average until the fifth month of life.

Among the causes of neonatal nasal obstruction, we recense: choanal atresia, pyriform aperture stenosis, craniofacial anomalies, meningo-encephalcele, dacrycystocele, tumor processes, traumatic lesions and Infectious rhinitis (gonococcus, chlamydia...) [3].

Nasal vestibular stenosis is an acquired process. In the literature, it has been described as a result of previous nasal surgical procedures, nasal packing, and excessive cautery for epistaxis, birth trauma, and flash burn injury [4-8].

Nasal vestibular stenosis may be also a rare consequence of prolonged instrumentation of the nasal cavity. It has been described initially as a complication of nasotracheal intubation.

Since its development in 1975, nasal CPAP is gaining favor as a method of respiratory support at several NICU [2]. It’s indicated for respiratory distress syndrome, patent ductus arteriosus, pulmonary edema, apnea, bronchiolitis or bronchopulmonary dysplasia, tracheomalacia and a lot of other neonatal respiratory distress syndrome [2]. As well as, it reduces the incidence of subsequent subglottic and tracheal stenosis, secondary infections and aspiration, which are associated with chronic intubation [9].

In a retrospective study from British Columbia, the use of nasal CPAP increased from 60% from 1996 to 2000 to 71% from 2000 to 2004 [10]. Similarly Jatana, et al. [11] have seen the same trend of nasal CPAP use in their NICU rising from 30% to 47% from 2004 to 2010.

Consequently, nasal deformities resulting from the use of the CPAP nasal prongs are increasingly described and individualized. Loftus, et al. [2] reported 8 patients with nasal deformities attributed to nasal CPAP use, including but not limited to nasal vestibular stenosis. De Rowe, et al. [12] described a 6-week-old patient with bilateral nasal synechia after 3 weeks of nasal CPAP use having been treated by endoscopic synechialysis, bilateral stenting, and repeated dilations. Smith and Roy [13] described two patients with vestibular stenosis after extended use of nasal CPAP. The first case described was a 4-month-old infant ex- 26 week premature infant, who had bilateral 80% nasal vestibular stenosis and significant cosmetic deformity. The second patient was a 5-month-old ex-25 week premature infant who had bilateral 95% nasal vestibular stenosis. Both corrected with Endoscopic lysis of nasal synechiae, nasal stenting and application of Mitomycin C. Similarly, Jatana, et al. [11] described a 7-week-old infant ex 29 week premature infant with bilateral nearly complete (>95%) vestibular stenosis at the tip where the nasal CPAP would have been positioned repaired by endoscopic lysis, application of mitomycin c, and nasal stenting. Our observation joins these cases, since it is a 3 week-old ex 31 premature infant with 80% bilateral nasal synechia after two weeks of nasal CPAP.

The incidence of intra-nasal synechiae is not well established, since isolated cases are described in the literature. Jatana, et al. [11], have conducted a cross-sectional study of 100 patients to characterize and determine the incidence of complications caused by nasal CPAP use in the NICU rather than to contrast nasal cannula with nasal CPAP. They have found an overall internal or external complication rate of 13.2%. Vestibular stenosis was seen in 4 nasal cavities (2.2%). Intrapasal complications were seen as early as 8 to 9 days after nasal CPAP administration. There have been no complications in the 9 patients with nasal cannula use alone.

iatrogenic nasal vestibular stenosis results from an insult to the nasal vestibular lining with resultant scarring and healing with contracture [14]. Jatana, et al. [11], have suggested the same pathophysiological mechanism previously described by minnigerode, et al. [15], involving pressure necrosis from the nasal prongs, air trauma itself, or bacterial contamination with stasis at the prong-mucosal interface.

Endoscopy serves as a diagnostic and therapeutic tool. However, facial mass scan is important to evaluate additional anatomic anomalies.

Old treatment strategies that have been reported for repair of nasal vestibular stenosis are auricular composite grafting [15,16], full-thickness skin grafting with pyriform aperture enlargement, [16] vestibular labial mucosal grafting [15]. These techniques are increasingly neglected,
because of the difficulty of handling a small cavity in this case nasal vestibule. Furthermore, additional intranasal incisions can lead to increased scarring [13]. Endoscopic lysis, application of mitomycin c, and nasal stenting are the techniques that have gaining favor for repair of nasal vestibular stenosis [11,13]. In our case we opted for synchuiolysis and repeated dilatations and good results were obtained. De Rowe, et al. [12] has incriminated inappropriate dilatations in the formation of intranasal synechiae in three-fourth of their patients. But once synechia formed, they have preferred gentle dilatations as part of the treatment protocol in order to avoid stent placement.

In conclusion, the treatment of each neonate with INS was tailored to each case since there is no therapeutic consensus in the literature concerning this age group.

**Conclusion**

In front of an apnea, it is necessary to evoke a nasal obstruction. The verification of choanes' permeability is a systematic gesture to do at birth and do not hesitate to redo the slightest doubt. A normal facial mass scan should be supplemented by a nasal endoscopy if strong clinical suspicion especially after prolonged intra nasal instrumentation.

**References**


