Management of Bronchomalacia in Infants Post-Cardiac Surgery Using Synchronized Nasal DuoPAP: A Novel Technology

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ABSTRACT
Background: Tracheo‑bronchomalacia (TBM) is the weakness in the structural integrity of the cartilaginous ring and arch. It may occur in isolation with prematurity or secondarily in association with various congenital anomalies. Bronchomalacia is more commonly associated with congenital heart diseases. The conventional treatment options include positive pressure ventilation with or without tracheostomy, surgical correction of external compression and airway stenting.

Aim: To use “synchronized” nasal Dual positive airway pressure (DuoPAP), a non‑invasive mode of ventilation as an alternative treatment option for bronchomalacia to avoid complications associated with conventional treatment modalities.

Study Design: Prospective observational study conducted in Army Hospital Research and Referral from Jul 2019 to Dec 2020.

Material and Methods: We diagnosed seven cases of TBM post‑cardiac surgery at our institute, incidence of 4.2%. Four infants were diagnosed with left sided bronchomalacia, 2 were diagnosed with right sided bronchomalacia and one with tracheomalacia. Those infants were managed by “synchronized” nasal DuoPAP, a first in ventilation technology by Fabian Therapy Evolution ventilator (Acutronic, Switzerland).

Results: All seven infants showed significant improvement with synchronized nasal DuoPAP both clinically as well as radiologically. None of the infant required tracheostomy and discharged to home successfully.

Conclusion: The synchronized nasal DuoPAP is a low cost and effective treatment option for infants with TBM. It could be attributed to synchronization of the breaths leading to better tolerance and compliance in paediatric age group.

Keywords: Bronchomalacia, non‑invasive ventilation, synchronized DuoPAP, tracheomalacia

INTRODUCTION

About 13.7% cases of airway malacia is associated with congenital heart disease, especially bronchomalacia.[1] It is mainly due to extrinsic compression by dilated right atrium and pulmonary artery.[2] Congenital or primary tracheo‑bronchomalacia (TBM) could be found alone or along with genetic conditions that weaken the walls of the airway, while the acquired form may occur due to trauma, chronic inflammation, and/or prolonged compression of the airways by enlarged structure nearby and complications from medical procedures such as endotracheal intubation or surgical manipulation. Acquired TBM is generally caused by the degeneration (break down) of cartilage that typically...
supports the airways. In most cases of acquired TBM, the underlying cause could not be identified.\cite{3-5} Neonates and small infants remain prone to malacia even after repair and removal of any extrinsic compression. Hence the luminal dimensions must be preserved post therapy. The conventional treatment options for bronchomalacia are conservative therapy with prolonged positive pressure ventilation with or without tracheostomy, surgical intervention such as relief of external compression and pexy operations (Pulmonary arteriopexy/Aortopexy/Bronchopexy), and lastly airway stenting.\cite{6} The non-invasive ventilation is an alternate treatment option to overcome complications associated with conventional treatment options. But rarely infants tolerate nasal Dual positive airway pressure (DuoPAP) and most often it does not benefit due to poor compliance. We present a case series of seven infants diagnosed with tracheo/bronchomalacia post-cardiac surgery, being managed with “synchronized” nasal DuoPAP, a novel technology wherein synchronization helped in better tolerance of the nasal cannula with positive pressure.

**MATERIAL AND METHODS**

After approval of the Hospital Ethical Committee, a prospective observational study was conducted in a tertiary care hospital of the Armed Forces. The study period was between Jul 2019 and Dec 2020. Total seven infants with TBM post-cardiac surgery were managed with synchronized nasal DuoPAP after informed consent from the parents. The pre-op data collected includes age, gender, weight, body surface area (BSA), type of congenital heart disease and presence or absence of pre-op congestive cardiac failure (CCF) & pulmonary artery hypertension (PAH). The intra-op details include cardio pulmonary bypass (CPB) time, aortic cross-clamping time and type of surgery as mentioned in Table 1. Five infants presented TBM in association with ventricular septal defect (VSD), and two were associated with transposition of great arteries (TGA). All infants were operated for the respective congenital heart disease and managed with mechanical ventilation in the post-op period followed by high flow nasal cannula (AIRVO 2, Fisher & Paykel Healthcare limited, Auckland, New Zealand with Optiflow junior nasal cannula) post extubation as per the institutional protocol. All infants manifesting with airway collapse and requiring re-intubation were further evaluated with NCCT chest with dynamic PEEP to confirm the diagnosis of bronchomalacia. TBM was defined as an appearance of deformity and narrowing of the trachea’s cross-sectional area by, at least, more than 25% on expiration as seen and documented by NCCT.\cite{7} Figures 1 and 2 shows CT images of an infant diagnosed with left sided bronchomalacia. Those infants were then managed with continuous synchronized nasal DuoPAP by Fabian therapy evolution ventilator (Acutronic, Switzerland) using INSPIRE nCPAP generator and circuit with silencer and INSPIRE nCPAP mask of appropriate size as shown in Figure 3. The initial CPAP (continuous positive airway pressure) pressure and upper pressure level (P Duo) in ventilator were set at 05 and 12 mmHg, respectively, for all infants to prevent alveolar collapse. When the pressure requirements to keep the alveoli, patent reaches below 08 mmHg of upper pressure level, the infants were then taken on high flow nasal cannula. During the DuoPAP period, the infants were fed through orogastric tube of appropriate size. (The ethical approval and consent are required and the same is obtained from Institutional Ethical Committee (IEC) and patients NOK respectively. IEC dated 05 Jun 2019).

**RESULTS**

Between Jul 2019 and Dec 2020, we diagnosed seven cases of TBM post-cardiac surgery at our institute,
incidence of 4.2%. Four infants were diagnosed with left sided bronchomalacia, 2 were diagnosed with right sided bronchomalacia and one with tracheomalacia. The demographic and clinical profile of these patients are summarized in Table 1. Out of 7 infants, 6 were male and 1 was female. The mean age was 3.76 months. The mean mechanical ventilation time was 10.85 days and the mean DuoPAP time was 19.14 days. No pneumothorax, subcutaneous emphysema, nasal mucosal bleed or any other adverse event was noted with use of synchronised DuoPAP. All infants showed significant improvement with synchronized nasal DuoPAP both clinically as well as radiologically. Figures 4 and 5 are radiological evidence of complete collapse of left lung due to bronchomalacia and its recovery post DuoPAP ventilation. None of the patients showed agitation during the nasal DuoPAP therapy. Also, none of the infant required tracheostomy and were discharged to home successfully after a mean hospital stay of 33.7 days. All the patients were managed at home with Airway clearance techniques such as chest physiotherapy, bronchodilators and antibiotics as and when required. Also advised for monitoring oxygen saturation via pulse oximetry at least twice a day and review in paediatric outpatient department. We followed these patients for six months and found no respiratory symptoms or chest X-ray findings suggestive of recurrence.

DISCUSSION

Extubation failure occurs in 11% of neonates post-cardiac surgery and the congenital airway anomaly is an independent risk factor for extubation failure in such cases. Even a slight reduction of the small infant airway has a relatively large impact on airway resistance. Young airways remain prone to malacia even after repair and removal of any extrinsic compression, the antero-posterior diameter of infant glottis measures 7 mm and posterior transverse
diameter of 4 mm. Even 1 mm of mucosal edema reduces the cross-sectional area by 35%. Most of our cases had either enlarged LPAs compressing the Lt bronchus or MPA compressing trachea in one case. Superimposing would be the inflammation caused by surgical manipulation and/or weak cartilaginous support which could explain the right sided bronchomalacia. Although we did not further investigate as to cause of the bronchomalacia as we thought it to resolve with growth of the child as cartilage would gain strength. The symptoms of TBM include expiratory airway collapse which lead to stagnation of secretions, atelectasis, recurrent infections, hypoxia, hypercapnea, and respiratory failure. Treatment may only be needed if signs and symptoms are present, quality of life is impaired, and/or there is complete or near-complete collapse of the airway. It could be medical which includes optimization of ciliary clearance of secretions and treatment of infection till lumen enlarges. Depending on severity, surgical treatment options for TBM that continues to progress may include silicone and/or long-term stenting, tracheobronchoplasty or pexy procedures, CPAP, tracheostomy. The accurate diagnosis of airway malacia plays a vital role in planning effective treatment. Masters et al. described endoscopic features of airway malacia at different levels. The bronchomalacia can be easily diagnosed with the dynamic 3D Computed Tomography. The conventional treatment modalities for bronchomalacia have their own set of complications such as prolonged hospital stay, high failure rates especially in surgical procedure/stenting and secondary infections due to their invasive nature. Non-invasive ventilation is an alternate treatment option to overcome such complications. Aasebo et al. reported a case of bronchomalacia successfully treated by long-term non-invasive ventilation by using Bi-level CPAP mode. The paediatric patients generally don’t accept mask ventilation as well as high flow nasal cannula easily. The synchronization of nasal DuoPAP (a non-invasive mode of ventilation) is a new in ventilation technology by Acutronic. The synchronization improves the compliance and tolerance especially in paediatric age group. All seven infants in our case series well accepted the nasal DuoPAP even without sedation. The continuous PEEP prevents airway collapse and keeps the airway open, which is the main aim in the treatment of bronchomalacia. It is a highly effective and low risk treatment option for infants and children with respiratory distress due to bronchomalacia. It also offers an additional advantage of tolerance and better patient management in pediatric age group. The risk of developing maxilla-mandibular hypoplasia associated with regular use of mask is also eliminated in nasal DuoPAP. This case series is having few limitations, it’s a single centre study with suboptimal sample size. The diagnosis was made based only on clinical picture and 3D CT and no bronchoscopy was done for any of the patient. The infants were assessed based on clinical and radiological evidence during the treatment. Serial arterial blood gas analysis has not been analysed to see the effect of the treatment as few patients did not have arterial line in situ during prolonged ventilation periods. Since the technology of synchronization with the nasal DuoPAP has been introduced for the first time, there is no study on synchronised nasal DuoPAP published till date, it was not possible to compare the results.

CONCLUSION

Synchronisation of the nasal DuoPAP leads to better compliance in the paediatric age group. The synchronized nasal DuoPAP is an effective treatment option for infants with TBM post-cardiac surgery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Masters IB, Chang AB, Patterson I, Wainwright C, Buntain H, Dean BW, et al. Series of laryngomalacia, tracheomalacia, and
bronchomalacia disorders and their associations with other conditions in children. Pediatr Pulmonol 2002;34:189-95.
2. Berlinger NT, Long C, Foker J, Lucas RV Jr. Tracheobronchial compression in acyanotic congenital heart disease. Ann Otol Rhinol Laryngol 1983;92:387-90.
3. Carden KA, Boiselle PM, Waltz DA, Ernst A. Trachomalacia and tracheobronchomalacia in children and adults: An in-depth review. Chest 2005;127:984-1005.
4. Choi S, Claire Lawlor C, Rahbar R, Jennings R. Diagnosis, classification, and management of pediatric tracheobronchomalacia: A review. JAMA Otolaryngol Head Neck Surg 2019;145:265-75.
5. Ridge CA, O’donnell CR, Lee EY, Majid A, Boiselle PM. Tracheobronchomalacia: Current concepts and controversies. J Thorac Imaging 2011;26:278-89.
6. Pillai JB, Smith J, Hasan A, Spencer D. Review of pediatric airway malacia and its management, with emphasis on stenting. Eur J of Cardiothorac Surg 2005;27:35-44.
7. Rozycli HJ, Houten MLV, Elliott GR. Quantitative assessment of intrathoracic airway collapse in infants and children with tracheobronchomalacia. Pediatr Pulmonol 1996;21:241-5.
8. Hollinger P. Clinical aspects of congenital anomalies of the larynx, trachea, bronchi and esophagus. J Laryngol Otol 1961;75:1-44.
9. Abel V, Barac S, Rozmanic V, Vukas D, Drescik I, Abel Jr VA. Aortopexy and bronchopexy for the management of severe tracheomalacia and bronchomalacia. Pediatr Int 2003;45:104-6.
10. Leape LL, Longino LA. Infantile lobar emphysema. Pediatrics 1964;34:246-55.
11. Boiselle PM, Ernst A. State-of-the-art imaging of the central airways. Respiration 2003;70:383-94.
12. Choo EM, Seaman JC, Musani AI. Tracheomalacia/tracheobronchomalacia and hyperdynamic airway collapse. Immunol Allergy Clin North Am 2013;33:23-34.
13. Kheir F, Majid A. Tracheobronchomalacia and excessive dynamic airway collapse: Medical and surgical treatment. Semin Respir Crit Care Med 2018;39:667-73.
14. Valerie EP, Durrant AC, Forte V, Wales P, Chait P, Kim PC. A decade of using intraluminal tracheal/bronchial stents in the management of tracheomalacia and/or bronchomalacia: Is it better than aortopexy? J Pediatr Surg 2005;40:904-7.
15. Aaseboe K, Berstad AK, Skadberg BT. Noninvasive treatment of bronchomalacia, successful ventilation of a severely ill infant. Acta Paediatr 2007;96:310-2.