CASE REPORTS

Tracheo-bronchomalacia in infants

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INTRODUCTION

Tracheobronchomalacia (TBM) is the airway collapse in the trachea and bronchi which may be due to congenital causes or acquired causes. Tracheomalacia is the most common congenital tracheal abnormality with a reported incidence of 1 in 2,100 children[1]. Presentation may vary with a spectrum of nonspecific respiratory symptoms. Diagnosis is often challenging at the initial presentation. We present 5 cases of tracheobronchomalacia associated with rare congenital anomalies which needed cardio-thoracic interventions.

CASE PRESENTATION

First child was 3 months old. He underwent an arterial switch operation at birth. Following the surgery, he developed a respiratory tract infection. He needed ventilatory support and weaning off was difficult. On imaging Chest x ray revealed whiteout of bilateral upper zones of the chest and right lower zone suggestive of lower respiratory tract infection with trapped secretions. Flexible bronchoscopy was performed, and it was revealed that he was having secretions at segmental bronchial level mainly in right and left upper lobe segmental bronchial tree and right middle and lower lobe segmental bronchial tree with bronchomalacia. Following bronchoscopy his respiratory vitals improved with improvement of Chest X ray. He was planned for slow weaning off from ventilator switching to noninvasive ventilation from invasive ventilation which facilitate regaining autonomic functional stability of airway. Mechanical ventilation was switched from synchronized intermittent mandatory ventilation (SIMV) to non-invasive, continuous positive airway pressure (CPAP) and kept him on sedation to slowly acquire the airway patency and autonomic stability of the airway caliber. With a successful slow weaning process, he was stepped down to ward in 2 weeks time.

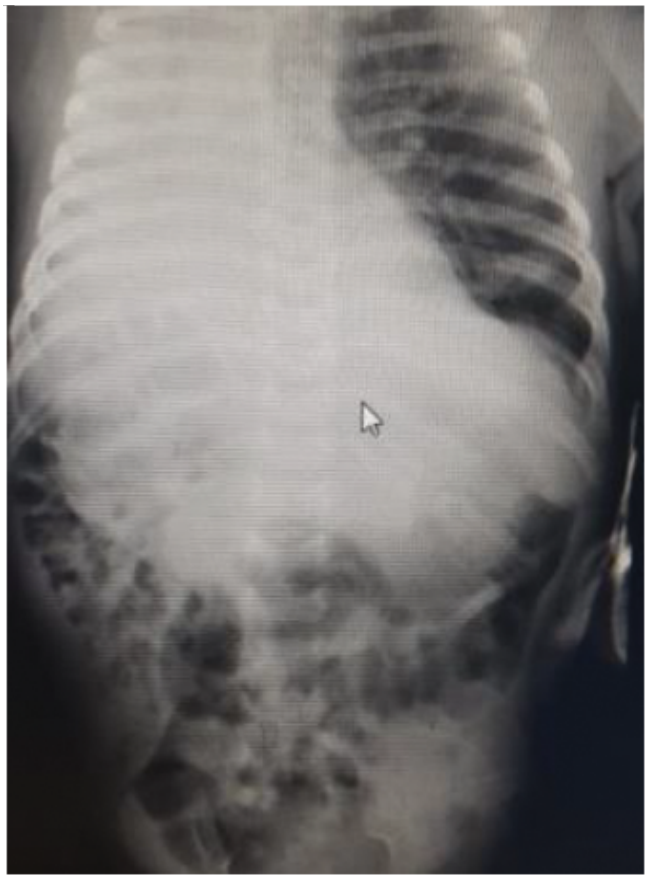

Figure 1. Right sided lung collapse with retained secretions.

The second child is 3 months old girl who underwent trans abdominal plication of right hemidiaphragm for diaphragmatic eventration. She was having respiratory distress since birth and was intubated and found to have right hemi diaphragmatic

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elevation. Following surgery, she was difficult to extubate and complicated with ventilator associated pneumonia where she was treated with antibiotics. Subsequent bronchoscopy revealed there is severe bronchomalacia involving from distal right main bronchus onwards up to segmental bronchial tree (Figure 1). Secretions were sucked out and she was planned for staged slow weaning from invasive ventilation to noninvasive ventilation (CPAP) to high flow. At the same time chest physiotherapy was continued and mother was encouraged to breast feed the child to facilitate the normal growth and development which will facilitate normal development of respiratory function along with autonomic functions of the respiratory tree.

Third case is another 3 months old child diagnosed with Di-George syndrome who had undergone truncus arteriosus repair and was in ITU for 2 months duration. Child had good recovery from cardiac surgery but had failed extubating 2 times since child developed respiratory distress following extubation. Flexible bronchoscopy was done and revealed to have tracheo-bronchomalacia involving distal trachea and at carina (Figure 2) and left main broncial-malacia up to lobar bronchi level (Figure 3). It was decided to wean off the child gradually with continued supportive care with chest physiotherapy and antibiotics to cover respiratory infection. Since the child was not responding to supportive therapy for two weeks it was decided to proceed with thoracotomy and posterior membranous tracheopexy. Due to ventilator associated pneumonia, the child went into sepsis and succumbed.

The other two cases are of innominate artery syndrome, which is a rare cause of tracheal compression in children. An eleven-month-old child was presented to the clinic with a history of difficulty in breathing congenital stridor and features of

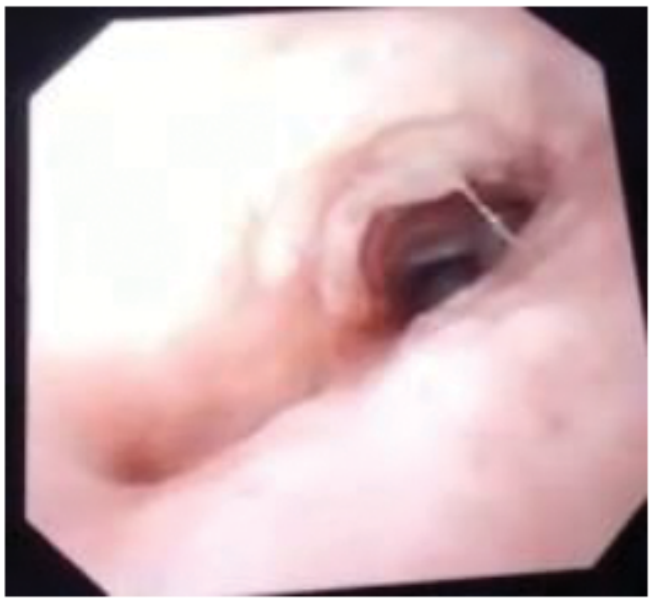

Figure 2. Tracheomalacia at carina.

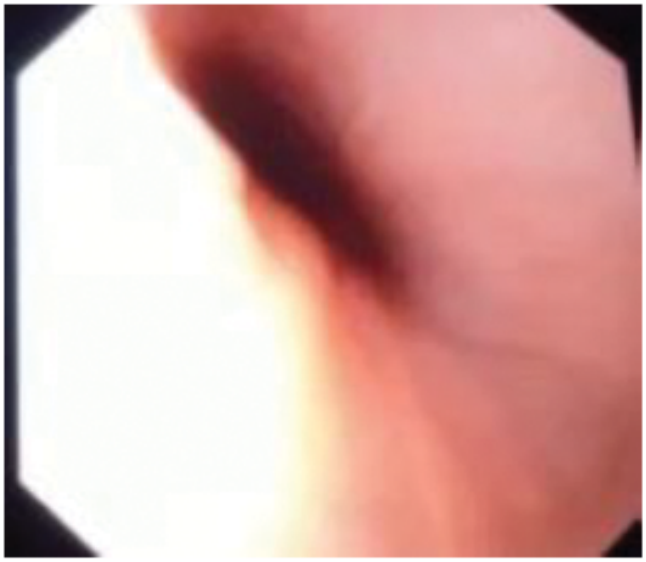

Figure 3. Opening of the distal left main bronchi.

VACTERL anomaly. He has undergone trachea oesophageal type C deformity and repair done along with colostomy for anal atresia and was referred for further management of his segmental tracheomalacia. On further evaluation his bronchoscopy revealed segmental tracheomalacia with pulsatile anterior tracheal compression at mid trachea (Figure 4) and CT angiogram revealed innominate artery compression of anterior trachea (Figure 5). Child has undergone angiography combined with broncho gram to assess the compression of trachea. It was decided to proceed with tracheopexy to relieve the focal tracheomalacia. Sternotomy was done and dissected the thymus tissue in anterior mediastinum and lymph node at pre-tracheal

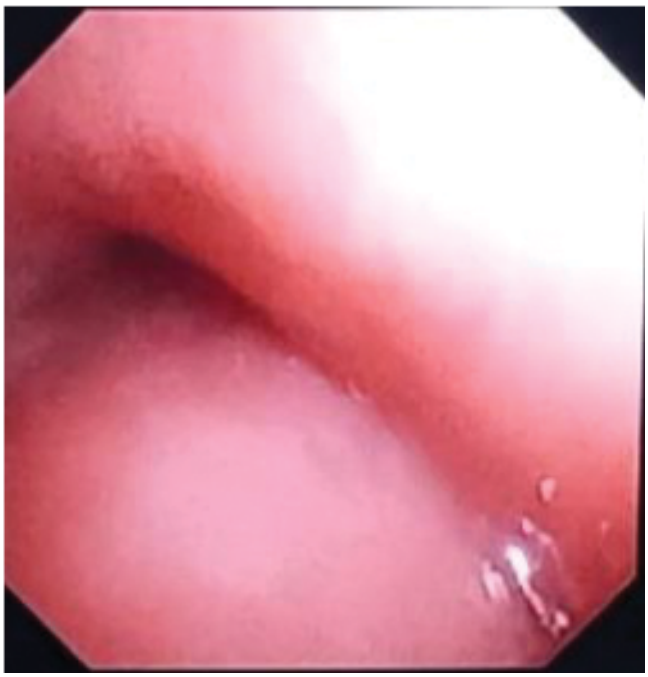

Figure 4. Focal tracheomalacia seen on fiberoptic bronchoscopy with complete dynamic collapse of tracheal lumen.

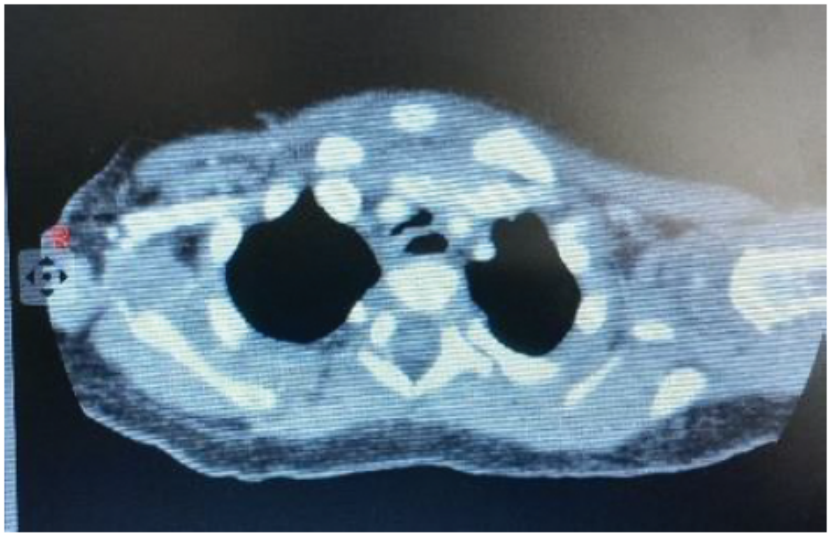

Figure 5. CECT chest showing innominate artery compressing trachea.

plane after slinging left bracheocephalic vein. There was no abnormal origin of the innominate artery and innominate arteriopexy done using polypropylene pledgeted sutures. On-table intra operative bronchoscopy revealed release of the anterior compression on trachea. It was decided to proceed with anterior tracheopexy to achieve optimal tracheal calibre and satisfactory tracheal lumen was demonstrated by intra operative bronchoscopy. The sternum was closed using polypropylene sutures and a reassessment of tracheal lumen was done following sternal closure.

A nine month old child presented with recurrent lower respiratory tract infections and failure to thrive. The child had previous history of tracheoesophageal fistula repair done at birth and undergone upper GI endoscopy which ruled out no recurrence of tracheoesophageal fistula. It was found that the child is having features of gastro-oesophageal reflux disease. For further evaluation, child has undergone aortogram and bronchogram which revealed child is having a bovine aortic arch with dynamic tracheal compression by innominate artery (Figure 6). It was decided to optimize her nutrition by PEG insertion and review her in clinic in one month and proceeding with flexible bronchoscopy to arrange surgical intervention for her tracheomalacia. On clinic review it was decided to proceed with elective surgery when she is 12 months of age.

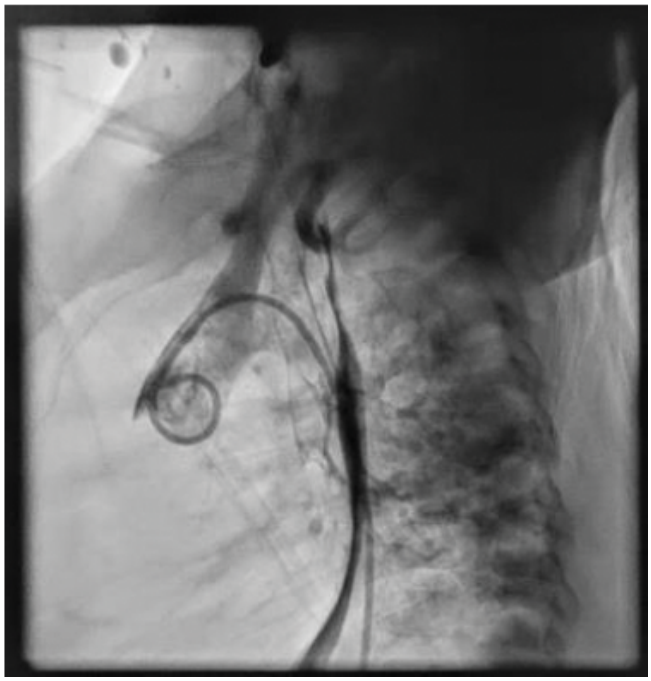

Figure 6. Contrast aortogram and bronchogram showing tracheal compression.

CONCLUSION

TBM often presents diagnostic challenges due to its varied clinical manifestations and the need for specialized tests such as dynamic imaging and bronchoscopy. It is often under-diagnosed or misdiagnosed due to its nonspecific respiratory symptoms. There are no definitive standardized guidelines for the evaluation, diagnosis, and treatment of TBM, which may lead to patients being initially misdiagnosed and incorrectly treated[2].

This case series highlights the association of tracheo-bronchomalacia with various congenital anomalies and the need for a multidisciplinary approach for proper evaluation, diagnosis, and treatment[3]. All these 5 cases were associated with surgeries done by cardiothoracic team. The above cases demonstrate the importance of considering tracheo-bronchomalacia in the differential diagnosis of respiratory distress, especially in patients with a history of congenital heart disease or other complex medical conditions.

It is important to evaluate suspected children with imaging and bronchoscopy, to accurately diagnose and manage the condition.

Key learning points

- TBM is commonly associated with other congenital pathologies where a child had to undergo corrective surgery (congenital heart diseases, tracheoesophageal fistula, congenital diaphragmatic hernia, congenital bowel pathologies etc.)
- This is common for surgeries where the child has been ventilated for long duration during post operative period and where associated cardiopulmonary pathologies and diseases of the chest wall are present (congenital cardiac diseases, lung hypoplasia, congenital diaphragmatic hernia and eventration).
- Tracheo-bronchomalacia with bronchopulmonary dysplasia leads to poor functional outcomes as it combines both anatomical and functional abnormalities of trachea bronchial tree and lung architecture.
- Slow weaning off from the ventilator from invasive ventilation to noninvasive ventilation with managing airway secretions by fibreoptic endoscopy and chest physiotherapy supported with treatment of ventilator associated pneumonia improves the outcome.
- Surgical correction is indicated in selective patients where aortopexy is done for severe tracheomalacia to allow more space for trachea at middle mediastinum in-between aorta and oesophagus. Surgical excision and anastomosis can be done in a limited number of patients with segmental involvement of the airway. These are associated with trachea-oesophageal fistula and atresia. There are techniques where the membranous wall of the trachea is plicated with mesh to reinforce the posterior wall in preventing collapse of the airway during respiration. Surgical outcomes are variable and depend on the expertise of the centre and the associated conditions of the child.

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