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Bilateral diaphragm paralysis after simultaneous cardiac surgery and Nuss procedure in the infant☆

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ABSTRACT

The case of a 15-month-old boy with bilateral diaphragm paralysis after simultaneous cardiac surgery for tetralogy of Fallot, and Nuss procedure for pectus excavatum, is presented. Extubated one day after his first operation, the boy suffered severe respiratory distress soon after, due to bilateral diaphragmatic paralysis. Diaphragm paralysis restricted abdominal respiration, while thoracic respiration was inhibited by metallic bar after the Nuss Procedure, which combined prevented extubation for 47 days. Thoracoplasty, such as the Nuss Procedure, should not be performed simultaneously with cardiac surgery because abdominal and thoracic respiration can be restricted in infants, causing prolonged, severe, post-surgical respiratory failure.

Bilateral diaphragm paralysis, a rare complication causing respiratory dysfunction in cardiac surgery in children, results in severe respiratory distress in infants or small children because abdominal respiration using the diaphragm is dominant [1]. Patients with bilateral diaphragm paralysis usually demonstrate thoracic respiration of small volume capacity. As thoracic respiration is more important in infant patients with bilateral diaphragm paralysis, restricting thoracic motion e.g. plastic surgery of the thorax, can cause further, severe, respiratory dysfunction causing prolonged mechanical ventilation. We report a case of severe respiratory distress resulting from simultaneous surgery of cardiac surgery and Nuss Procedure.

1. Case report

A boy, born at 38 weeks of gestation, was diagnosed with Pierre-Robin syndrome, tetralogy of Fallot and pectus excavatum. The Haller index of the boy was 3.32 at operation. However he had presented upper airway obstruction due to Pierre-Robin syndrome and mild tracheal stenosis was diagnosed with Bronchoscopy before surgery. Right ventricular outflow reconstruction had the risk of postoperative tracheal stenosis without aortopexy and thoracoplasty. Therefore cardiac surgery and thoracoplasty were simultaneously performed at 15 months. The cardiac surgery was a patch closure of a ventricular septal defect, right ventricular outflow reconstruction and aortopexy by median sternotomy, while the thoracoplasty was a Nuss procedure also under direct vision. A short and straight shaped bar was placed under the anterior chest so that the bar could be removed easily. After surgery, continuous intravenous fentanyl was administrated for postoperative pain control. At one day after surgery, severe respiratory distress was recognized soon after extubation, despite no apparent upper airway obstruction; circulatory failure was recognized before bilateral diaphragmatic paralysis was diagnosed in fluoroscopy (Fig. 1). A bilateral diaphragm plication was performed 5 days after the first operation, soon free of mechanical ventilation.

However, respiratory distress continued even after the plication, resulting in difficulty weaning the patient from mechanical ventilation. His crying vital capacity was only 5.1 ml/kg (while normal value is 15 ml/kg +). We considered that severe respiratory failure may have resulted from cardiac failure after cardiac surgery plus the restriction of thoracic respiration influenced by Nuss procedure, in addition to bilateral diaphragmatic paralysis.

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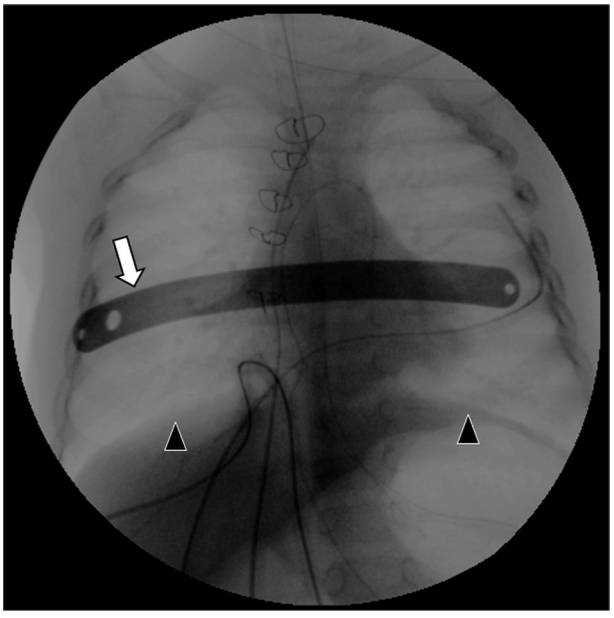

Fig. 1. Bilateral diaphragm paralysis in fluoroscopy. Spontaneous respiratory motion is unclear because Nuss bar (arrow) restricts thoracic respiration and diaphragm is paralyzed at the level of the eighth rib in either side (arrowheads).

Respiratory rehabilitation and trans-intestinal nutrition were planned to improve his respiration. Nasoduodenal nutrition and physiological rehabilitation, including chest physiotherapy, were introduced with tapering sedatives. At 47 days from the first operation, the boy was extubated, followed by non-invasive bi-level positive airway pressure ventilation using a nasal mask to support spontaneous respiration (Respironics, Pittsburgh, PA, USA). Non-invasive respiratory support was given for 5 days. The boy was discharged home at 54 days after his cardiac surgery.

2. Discussion

Bilateral diaphragm paralysis is a rare complication causing respiratory distress while unilateral diaphragm paralysis is less severe than bilateral diaphragm paralysis [2,3]. Ovadia et al. described that the incidence of bilateral diaphragm paralysis after pediatric cardiac surgery is 0.28% [1], although Maaike de Leeuw et al. reported that incidence of unilateral diaphragm paralysis after pediatric cardiac surgery is 1.6% [2]. Diaphragm paralysis normally requires mechanical ventilation for long periods due to respiratory distress, suggesting more symptomatic respiratory failure in infants or small children as abdominal respiration is dominant. Thus, bilateral diaphragm paralysis can cause particularly critical situations in infants, impairing respiratory function resulting in chronic respiratory failure. Bilateral diaphragm paralysis is a rare post-cardiac surgery complication; severe respiratory failure requires much more careful management than unilateral diaphragm paralysis.

Nuss Procedure obtains several clinical issues, while it is well-recognized as a useful surgery for funnel chest for children. Dyspnea and atelectasis are common post-operative complications of a Nuss Procedure, owing to restricted chest wall motion due to pain and metallic bar. Densmore et al. recommended a sufficient pain therapy example of epidural anesthesia during the early post-operative period [4]. Futagawa et al. also reported that continuous epidural infusion provides effective post-operative pain relief as well as preventing complications [5]. In this case, however, it was impossible to use epidural anesthesia on account of the boy's small body, and his heparinization for cardiac surgery. Therefore the boy had severe chest pain, causing severe respiratory failure immediately after Nuss Procedure. Clearly, pain control is vital to maintain sufficient ventilation after Nuss Procedure.

Diaphragmatic plication is a useful procedure for phrenic nerve palsy after cardiac surgery in infants. Illze et al. reported early plication substantially reduces the duration of ventilation, with the associated reduced morbidity and ICU stay [6]. Lemmer et al. reported the effects of diaphragmatic plication for post-surgical diaphragmatic paralysis concluding that diaphragmatic plication carries low mid-term risk in terms of recovery of phrenic nerve function, lung function values and exercise capacity [3]. Therefore, early-stage diaphragmatic plication is particularly suitable for infants who have severe respiratory distress.

There are several reports describing the usefulness of Non invasive positive pressure ventilation (NIPPV) for bilateral diaphragm paralysis after cardiac surgery [7,8]. NIPPV supports spontaneous respiration non-invasively, resulting in the avoidance of prolonged tracheal intubation and tracheostomy. In contrast, some reports mention that conservative management is efficient for treatment against bilateral diaphragm paralysis. Ovadia et al. described nine cases of bilateral diaphragm paralysis after pediatric cardiac surgery which were managed with mechanical ventilation, resulting in extubation in less than 7 weeks without tracheostomy [1]. As our patient presented severe respiratory distress after extubation, he urgently required an alternative means of respiratory control. If the patient's respiratory distress is so severe that it is difficult to control respiration only by NIPPV, conservative care (including tracheal intubation) may be appropriate.

Our plan was to combine cardiac with plastic surgery, so that only one anesthesia was required, and that the risk of mediastinal damage by Nuss bar after thoracotomy would be reduced. Unfortunately, unexpected complications caused by simultaneous restriction of abdominal and thoracic respiration transpired. Severely impaired respiratory function was caused by combined complications from each surgery, although these respiratory restrictions occasionally happen in either surgery. Thus, cardiac surgery and thoracoplasty should not be performed simultaneously if the advantages of simultaneous surgery do not significantly outweigh the disadvantages, as extraordinary complications in simultaneous surgery can occur.

3. Conclusion

Cardiac surgery and thoracoplasty should be performed separately, in order to avoid simultaneous respiratory complications in each procedure which can result in greater risk for severe respiratory failure after the surgeries.

Conflict of interest

The authors declare no conflict of interest associated with this manuscript.

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