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Indian Journal of Clinical Anaesthesia

Journal homepage: www.ijca.in



Brief Communication

A rare case of congenital bilateral bullous emphysema with vacterl anomaly with congenital heart disease

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ARTICLE INFO

Article history: Received 12-04-2021 Accepted 21-04-2021 Available online 11-11-2021 This is an Open Access (OA) journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

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Dear editor,

Congenital bullous emphysema is defined as large bullae involving one third of hemithorax. A 2yr, 9 months old female weighing 8.2kg presented with dyspnea on playing was posted for surgical correction of congenital heart disease. She is a case of VACTERL anomaly having polydactyly, vertebral anomalies and single right kidney, her developmental milestones was delayed and immunisation was complete. On 2D echocardiography she was diagnosed with ventricular septal defect with multiple atrial septal defect with severe pulmonary arterial hypertension. Routine investigation were within normal limits. Chest radiography showed right sided pneumothorax so we decided to insert intercostal drain and proceed with diagnostic catheter study for evaluation for severe pulmonary arterial hypertension. Chest radiography revealed no reduction in pneumothorax so we did high resolution computed tomography which showed bilateral emphysema involving middle and lower lobe of both lung (Figure 1). The radiographic criteria for congenital bullous emphysema as defined by Roberts and colleagues, include the presence of giant bullae in one or both upper lobe, middle lobe, lower lobe, occupying atleast one third of hemithorax and compressing surrounding normal lung parenchyma.² Differential diagnosis include bronchial atresia, bronchogenic cyst, Swyer-james syndrome and congenital cystic adematoid

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malformation. Surgical removal of affected lobe is commonly done. We do conservatively manage some patients who are not clinically in respiratory distress and able to feed and grow. Maintaining ventilator pressures and volume as low as possible avoids producing ventilator related hyper-expansion of the affected lobe. Management by more conservative, gentle ventilation techniques if successful will result in fewer emergency surgeries with congenital lobar emphysema.³

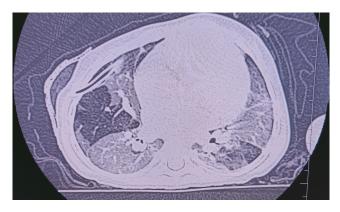


Fig. 1: High resolution computed tomography showing bullous changes and progressive emphysema and intercostal drain in position

For performing emergency lobectomy the anaesthetic management would be a challenge. After induction, PEEP

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can expand the affected lobe and this can compress the normal lung resulting in cardiovascular compromise. Once resection of the affected lobe is completed controlled lung ventilation with muscle relaxation should be done. Blood gas monitoring intraoperatively and postoperatively should be done. Intercostal block is a good for pain control. Lung biopsy of the resected part should be done.

Conflicts of Interest

There are no conflicts of interest.

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Cite this article: Jadhav P, Thorat M, Shetty N. A rare case of congenital bilateral bullous emphysema with vacterl anomaly with congenital heart disease. *Indian J Clin Anaesth* 2021;8(4):626-627.