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Case Report

Burst or Bluff: A Bulla Masquerading as Tension Pneumothorax in a 2-Year-Old Child

Anurag Goyal[1*], Swarna Poundrik[2], Chiranjeeb DebBarma[3], Ramakant Dixit[2], Kanwar Singh[4], Neeraj Gupta[2]

[1]Department of Cardiovascular & Thoracic Surgery, J L N Medical College, Ajmer, Rajasthan, India
[2]Department of Respiratory Medicine, J L N Medical College, Ajmer, Rajasthan, India
[3]Department of General Surgery, J L N Medical College, Ajmer, Rajasthan, India
[4]Department of Paediatrics, J L N Medical College, Ajmer, Rajasthan, India

Abstract

This case highlights the diagnostic challenge of differentiating between a pneumothorax and a congenital pulmonary bulla, particularly in a malnourished paediatric patient presenting with chronic respiratory symptoms. Despite initial management with intercostal drainage under the impression of pneumothorax, the absence of lung expansion prompted further imaging, which confirmed congenital bullous disease. Definitive surgical management with an 80 mm linear stapler resection resulted in marked clinical improvement. Early consideration of bullous lung disease in children with atypical or non-resolving pneumothorax is essential to avoid unnecessary interventions and to ensure timely surgical correction for favourable outcomes.

Keywords: Giant bullae, Pneumothorax, Linear Stapler Resection.

Introduction

A bulla is defined as an airspace measuring greater than 1-cm, which is sharply demarcated by a thin wall (≤1-mm in thickness). They are subdivided further into 3 types: Type I, a small amount of hyperinflated lung tissue that is narrow (pedunculated) and contains no lung parenchyma; Type II, a relatively smaller amount of hyperinflated lung tissue that is broad (sessile); and Type III, a large amount of hyperinflated lung tissue extending to the pulmonary hilum, with ill-defined margins and minimal parenchyma in each bulla.[1] Another way to classify bullae is the Klingman classification[2] of bullae, consisting of 2 types of bullae The first type is a bullae with structurally normal surrounding lung tissue, which is seen in about 20% of patients. This is an isolated bulla without generalized emphysema. It is associated with congenital bullous disease. In such cases, surgical resection usually has a good prognosis. The second type is a bullae with abnormal surrounding lung tissue, seen in about 80% of patients, which is associated with emphysema, COPD, or diffuse lung disease (acquired).

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The aetiology of congenital bulla traces to abnormal alveolar development or bronchial atresia. It can also be associated with congenital lobar emphysema, Marfan's syndrome, Ehlers–Danlos syndrome, and α1-antitrypsin deficiency. It is often localised and may remain asymptomatic until complicated, i.e., rupture leading to pneumothorax or progressive enlargement. Acquired bulla develops secondary to destruction of alveolar walls later in life, like in cases of smoking-related emphysema, it can also occur post infection sequalae, like in tuberculosis or bacterial pneumonia, trauma, COPD, silicosis, HIV related pneumocystis infection, etc.[3,4]

Large pulmonary bullae, when seen on chest X-ray, may resemble a pneumothorax due to their radiolucency and absence of lung markings.[5] These can be differentiated with the help of radiological cues. In bullae, there is a thin curvilinear wall and are usually oval or rounded in shape,

Address for correspondence: Anurag Goyal
Department of Cardiovascular & Thoracic Surgery, J L N Medical College, Ajmer, Rajasthan, India
E-mail: anuragaiimsctvs@gmail.com

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and sometimes internal septations can also be seen. At the same time, in a pneumothorax, there is an absence of lung markings peripheral to the pleural line with no visible thin wall. CT remains the gold standard in distinguishing between the two.[6] Misinterpretation in such cases can lead to erroneous intercostal chest tube insertion, risking entry into the bulla, which can then cause serious complications such as bronchopleural fistula, haemorrhage, or infection. Hence, giant bullae should be included in the differential diagnosis of a pneumothorax and should be ruled out before the placement of a chest tube. Here, we are presenting such a case where a giant bulla being misdiagnosed as a pneumothorax led to the erroneous placement of a chest tube in a 2-year-old child, a very rare presentation in a child of such a young age.

Case Report

A 2-year-old male child with malnourishment (weight 50% of normal weight for age) presented to the emergency department with complaints of shortness of breath for 5 months, more from the last 2 days, along with decreased appetite for 1 month, and fever, cough with cold for 3 days. On examination, the patient was drowsy, irritable and lethargic with a respiratory rate of 48/min, pulse rate 146/min, SpO2 87% at room air. Patient was afebrile. A high flow nasal cannula was attached & SpO2 of 90% was achieved. On respiratory system examination, tachypnoea along with intercostal and subcostal recessions were noted. A hyper-resonant note was felt on percussion over the entire right side of the chest and the intensity of breath sounds was found to decrease over all of the right side on auscultation. Routine blood investigations revealed haemoglobin 9.1 gm/dl, total leukocyte count 10500 cells/mm^3, haematocrit 26.60, platelets 587 thousand/µL, urea 18.1 mg/dl, creatinine 0.5mg/dl, Na 135.5 meq/l, K 4.3 meq/l, Cl 90.9 meq/l and raised quantitative CRP (40.1 mg/dl). Viral markers were negative. HRCT chest was done prior to admission of the patient to our centre, which was suggestive of a large right-sided pneumothorax with right collapsed lung and mediastinal shifting to the contralateral side (Figure 1). An intercostal chest tube drain (ICTD) was inserted in the right midaxillary line in the 5^{th} intercostal space by the attending department. Air leak and water column movements were present post-ICDT insertion and on chest x-ray, the lung did not expand (Figure 2).

On day 2 post ICDT Insertion, the ICDT was readjusted, but the lung did not expand, with no relief in dyspnoea, and intercostal and subcostal recessions were still seen. On day 3 post-ICDT, column movements became absent and the intensity of breath sound was decreased on the right side on auscultation with worsening dyspnoea. A thoracic surgical opinion was taken, and a CECT Chest was advised, which was suggestive of congenital bullous disease (Figure 3).

Surgical exploration was decided in view of the patient's condition and respiratory distress. The patient underwent right posterolateral thoracotomy and a large bulla was found (Figure 4). It was managed by 80 mm linear stapler resection and chest tube placement (Figures 5 and 6). Patient was ventilated electively overnight and extubated the next morning. Vitals were now stable and the patient's condition drastically improved after intervention. Post procedure chest X-ray of the patient showed completely expanded lung with ICDT in situ (Figure 7).

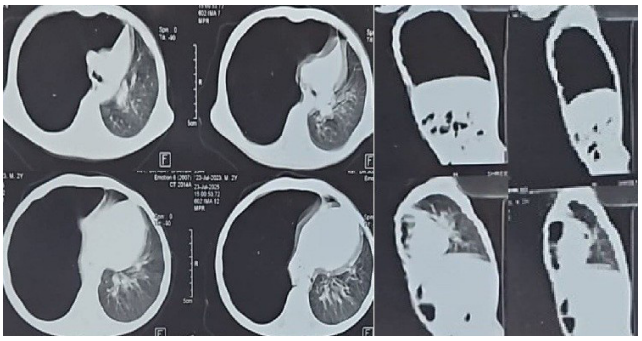


Figure 1: HRCT image suggesting a large right-sided pneumothorax causing severe compression on the left lung and trachea-mediastinal shift to the left

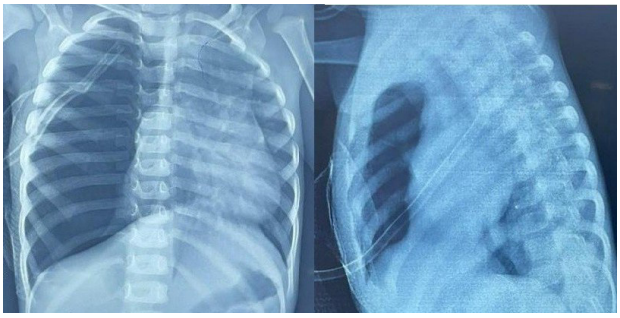


Figure 2: Chest X-ray image of the patient post ICDT placement showing hyperlucency at the right hemithorax, suggestive of un-expanded lung even after ICDT insertion. Mediastinal shift to the contralateral side is also evident

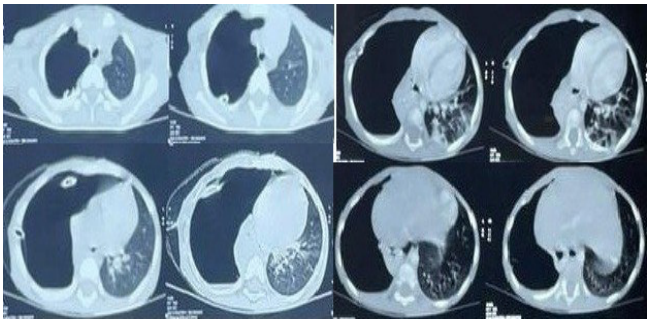


Figure 3: CECT chest of this patient is suggestive of large bullae on the right side with the mediastinum towards the left side. Multiple thin septations can also be appreciated, along with the ICDT placed inside the large bullae



Figure 4: Intraoperative image of the giant bullae that were found after postero-lateral thoracotomy

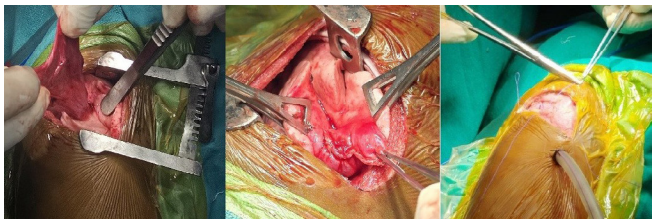


Figure 5: Intra-operative images of linear stapler resection of the giant bullae in sequence



Figure 6: 80 mm linear stapler

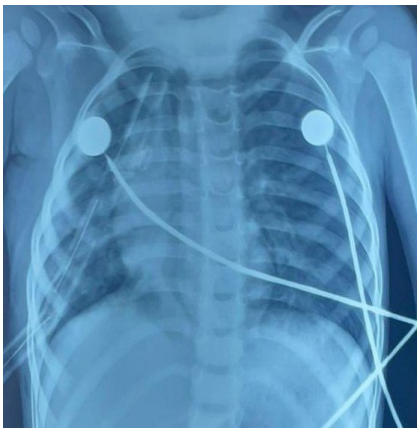


Figure 7: Chest X-ray of a patient with ICD showing expanded lung

Discussion

This case illustrates the need for prompt and accurate imaging choices to be made by the treating doctor in such cases where pneumothorax can mimic a bulla. The gold standard method to distinguish a bullae form pneumothorax is CECT Chest. However, in some cases, there can be a giant-bullae with a co-existing pneumothorax. That can also be distinguished on CT chest with the help of a radiological sign called the "double wall sign."[7] It is characterised by air outlining both the inner and outer walls of a bulla or cyst, making its wall appear "double." It occurs when a pneumothorax develops adjacent to a pulmonary bulla. Air in the pleural space outlines the outer surface of the bulla wall, while air inside the bulla outlines the inner surface. It is highly suggestive of the coexistence of bulla and pneumothorax. If double wall sign is absent on thoracic CECT, it goes against the diagnosis of a pneumothorax. The limitation of the 'Double Wall Sign' is that sometimes it can be mimicked by two adjacent bullae, leading to a false double wall sign. This can be avoided by careful selection of contiguous CT slides, apart from utilizing history and physical examination.[8]

The prognosis of bullous disease depends on the size, location, and number of bullae, as well as the condition of the surrounding lung parenchyma.[9] An isolated congenital bulla with otherwise normal lung tissue generally has an excellent prognosis after surgical resection. But in diffuse bullous disease or association with syndromes (e.g., Marfan's, Ehlers–Danlos, α1-antitrypsin deficiency), prognosis is poor. Untreated large bullae may cause recurrent pneumothorax, infection, and progressive respiratory compromise, leading to poor outcomes. Most children show improvement in symptoms and lung function, as seen in this particular case.[10] If resection is complete, there are low chances of recurrence. Malnutrition or comorbidities (as in this case) may worsen perioperative risks, but the long-term outcome is favorable if the lung functions are preserved.

Bullae being misdiagnosed as a pneumothorax is a common presentation in the adult population, as was found in available literature.[11-14] However, a similar presentation in a child of such a younger age group has hardly reported, which makes this case a unique one and intrigues academic interests.

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