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Dual Foregut Duplications in a Neonate: A Case Report of Bronchogenic and Esophageal Duplication Cysts in the Same Individual

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Abstract

Esophageal duplication cysts and bronchogenic cysts are rare congenital anomalies of the foregut that typically develop as distinct entities. Their coexistence within a single lesion represents an exceptionally rare developmental anomaly, arising from aberrant differentiation of both the dorsal and ventral components of the primitive foregut. To the best of our knowledge, this report is among the first to describe a case of esophageal duplication cyst with an associated bronchogenic cyst in a neonate. While Jain et al. (2017) previously documented a similar coexistence, our case is unique due to the early onset of severe respiratory distress necessitating prompt surgical intervention. This case underscores the critical need for clinicians to consider foregut duplication cysts in neonates with unexplained respiratory distress and highlights the importance of early diagnosis and timely surgical management in preventing life-threatening complications.

Keywords: Esophageal duplication cyst, Bronchogenic cyst, Neonatal respiratory distress, Foregut malformation, Surgical excision

Case Presentation

A full-term male neonate, with antenatally detected multiple intrathoracic paraspinal cystic lesions, born at 38 weeks of gestation via lower segment caesarean section (LSCS) due to polyhydramnios, presented with severe respiratory distress immediately after birth, necessitating urgent intubation. The infant was gradually weaned off ventilatory support and extubated on day 7 of life. However, he developed worsening respiratory

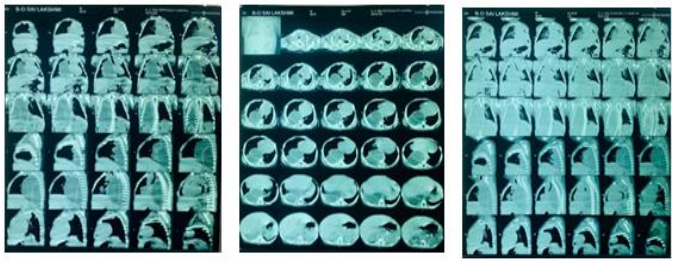
distress with desaturation, requiring re-intubation. Due to persistent respiratory compromise, he was referred to our institution for further evaluation.

On admission, the neonate was tachypneic and required synchronized intermittent mandatory ventilation (SIMV) for respiratory support. Systemic examination revealed decreased bilateral air entry and a downward displacement of the liver, suggesting a significant mass effect in the thoracic cavity.

Investigations

A chest X-ray (Figure 1) demonstrated a homogeneous opacity in the right lung, accompanied by a contralateral mediastinal shift and compression of the left lung. A chest ultrasonography identified a well-defined cystic mass measuring 6x3.1 cm in the right paraspinal thoracic region, along with additional complex cystic lesions and minimal right pleural effusion. Computed Tomography (CT) of the Thorax (Figures 2, 3 and 4) revealed a multiloculated cystic lesion extending from the right clavicle to the posterior renal region, contributing to a contralateral mediastinal shift and minimal right pleural effusion. Based on these findings and the progressive respiratory compromise, the decision was made to proceed with surgical excision of the cystic lesion.

Figure 1: Chest X-ray showing a homogeneous opacity in the right lung with contralateral mediastinal shift



Figures 2, 3 and 4: CT thorax images demonstrating multiple cystic lesions causing mass effect and mediastinal shift

Management

A right posterolateral thoracotomy was performed (Figure 5). Intraoperatively, a thickened parietal pleura with necrotic tissue was observed. A large right lower lobe cyst arising from the bronchus was excised, along with a posterior mediastinal cyst measuring 7x5 cm, containing mucosa and muscular layers.

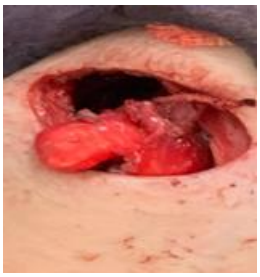


Figure 5: Intraoperative image showing the cystic mass during surgical excision.

Histopathological examination revealed a cyst wall lined by tall columnar mucin-secreting epithelium, arranged in villi and crypts. The cyst also exhibited a bi-layered hypertrophied muscular wall with submucosal edema and congested vessels. Additionally, there was evidence of granulation tissue response, along with surface ulceration, necrosis, and hat mosiderin-laden macrophage collection. These findings were consistent with an esophageal duplication cyst associated with a bronchogenic cyst [1].

Postoperatively, the neonate remained on SIMV and was successfully extubated on post-operative day 5. The

post-operative course was complicated by pneumothorax and sepsis, which were managed with intercostal drainage (ICD), intravenous antibiotics, and other supportive care measures. The neonate made a full recovery and was discharged on day 31 of life with stable respiratory function.

At the time of writing this article, the child is 1 year 3 month old and is thriving well.

Discussion

Foregut duplication cysts, including esophageal duplication and bronchogenic cysts, constitute a significant proportion of mediastinal masses in neonates and children [2-3]. While typically occurring as separate entities, their coexistence has been rarely reported in the literature [1]. These cysts arise due to aberrant embryonic foregut development, involving both the dorsal and ventral aspects of the primitive foregut [4].

In a study by Kawashima et al., a newborn with a cervical esophageal duplication cyst presented with progressive airway compression, requiring early surgical excision, similar to present case[5]. A close relationship has been found between esophageal duplication cysts and bronchogenic cysts based on several studies.[5] Other congenital anomalies can be associated with esophageal duplication cysts such as duplications of intestines, spinal anomalies etc.[6]

Imaging techniques, including ultrasound, CT, and MRI, are critical in diagnosing these cysts, and complete surgical excision remains the gold standard for treatment [7-8]. There is a risk for hemorrhage, infection or malignant transformation in these cysts.[9] This highlights the importance of early detection and intervention. The necessity for surgical excision has been emphasized in multiple studies, as untreated cases may lead to complication [1,2]. Our patient benefited from early surgical management, leading to a favorable outcome.

To further contextualize the rarity of this case and compare it with previously reported instances, we summarize key findings from relevant studies in the literature, as presented in the following review table.

Conclusion

The coexistence of esophageal duplication cysts and bronchogenic cysts is rare but should be considered in neonates presenting with unexplained respiratory distress. Early diagnosis and surgical excision are crucial to prevent complications and optimize outcomes. This case emphasizes the importance of a multidisciplinary approach, incorporating radiological assessment, prompt surgical intervention and meticulous postoperative care in managing such congenital anomalies [2].

Author	Year	Findings	Management	Outcome
Kawashima et al [7].	2016	Cervical esophageal duplication cyst causing airway obstruction	Early surgical intervention	Recovery without complications
Jain et al [1].	2017	Coexistence of bronchogenic and esophageal duplication cysts	Surgical excision	Favorable
Wahi & Fernando [2]	2023	Review of esophageal duplication cysts	Surgical excision recommended	Favorable outcomes in most cases
Gross et al [4].	2023	Bronchogenic cysts and their clinical presentation	Imaging-based diagnosis and surgical excision	Favorable

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