Journal of Academic Research
Vol 30, Issu Special issue, 2026 "16-22"

Congenital neuroentric cyst and Synchronous thoraco-abdominal enteric duplication cysts in Misrata – Libya

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Article information

Key words
Neurenteric cyst, Duplication cyst, ectopic gastric mucosa, thoracotomy

Received: 11-09-2025
Accepted: 03-11-2025
Available: 01-01-2026

Abstract

Background: Neurenteric cysts are rare congenital anomalies arising from abnormal separation of the embryonic notochord and foregut. Their association with midgut enteric duplication is exceptionally uncommon. Enteric duplication cysts (EDCs) themselves are rare developmental anomalies that may occur anywhere along the gastrointestinal tract, demonstrating wide variability in size, location, and symptoms. This study reports a rare case of a neurenteric cyst associated with midgut duplication in a Libyan patient and highlights its embryological background, diagnostic challenges, and surgical management.
Methods: A comprehensive clinical, radiological, operative, and histopathological evaluation was performed. Diagnostic imaging included abdominal ultrasound and CT scan. Operative findings were documented, and histopathological analysis confirmed the nature of the lesion. A literature review was included to contextualize this unusual case.
Results: Radiological imaging demonstrated a cystic abdominal lesion consistent with an enteric duplication structure. Intraoperative exploration confirmed a neurenteric cyst closely associated with duplicated midgut loops. Complete excision was performed without complications. Histology showed gastrointestinal-type mucosa and features characteristic of neurenteric cysts. The patient had an uneventful recovery and remained asymptomatic on follow-up.
Conclusion: This case highlights the extreme rarity of neurenteric cysts associated with midgut duplication. Early recognition and surgical management are essential to avoid complications such as obstruction, infection, or perforation. This report contributes to the limited global and regional literature on complex EDC variants.

I) INTRODUCTION:

Enteric duplication cysts (EDCs) are uncommon congenital anomalies of unknown etiology{ 1,2,3 } The presentation of duplication cysts depends on their location, size and other factors such as the presence of the gastric mucosa.{4,5}

Neurenteric cysts are uncommon developmental anomalies arising from aberrant separation of the notochord and endoderm during embryogenesis. These cysts are often associated with other congenital anomalies, including vertebral, spinal cord, and gastrointestinal abnormalities. Midgut

duplication, another rare congenital condition, involves the presence of a duplicated segment of the midgut, often leading to significant clinical complications.

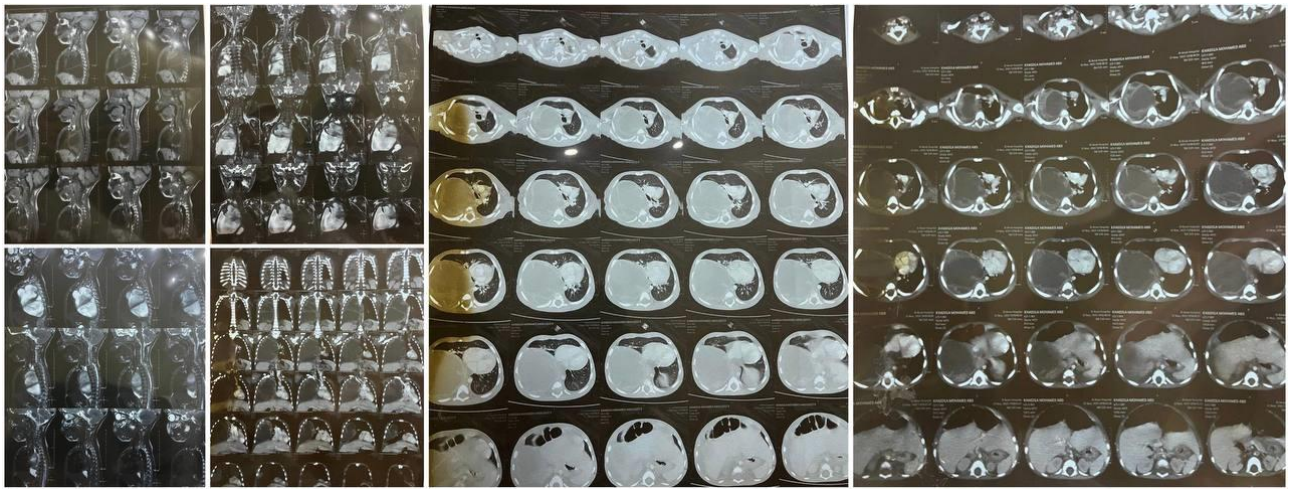
[3,4] Ectopic gastric mucosa is reported to be found in 20-30% of these duplications. {2,5,6} The ileum is the most common site for duplication of the alimentary tract whereas rectal, duodenal, gastric and thoraco-abdominal locations are extremely rare.[4] Synchronous multiple duplications are found in up to 15% of the cases.[4.5}

We present a case of a 5-month-old female infant diagnosed with a congenital neuroenteric cyst, leading to significant health challenges, including failure to thrive (FTT) and anemia.

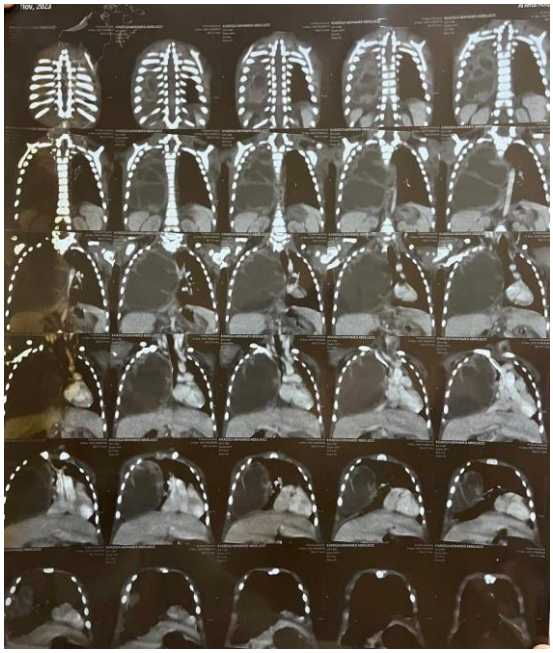
II) CASE REPORT

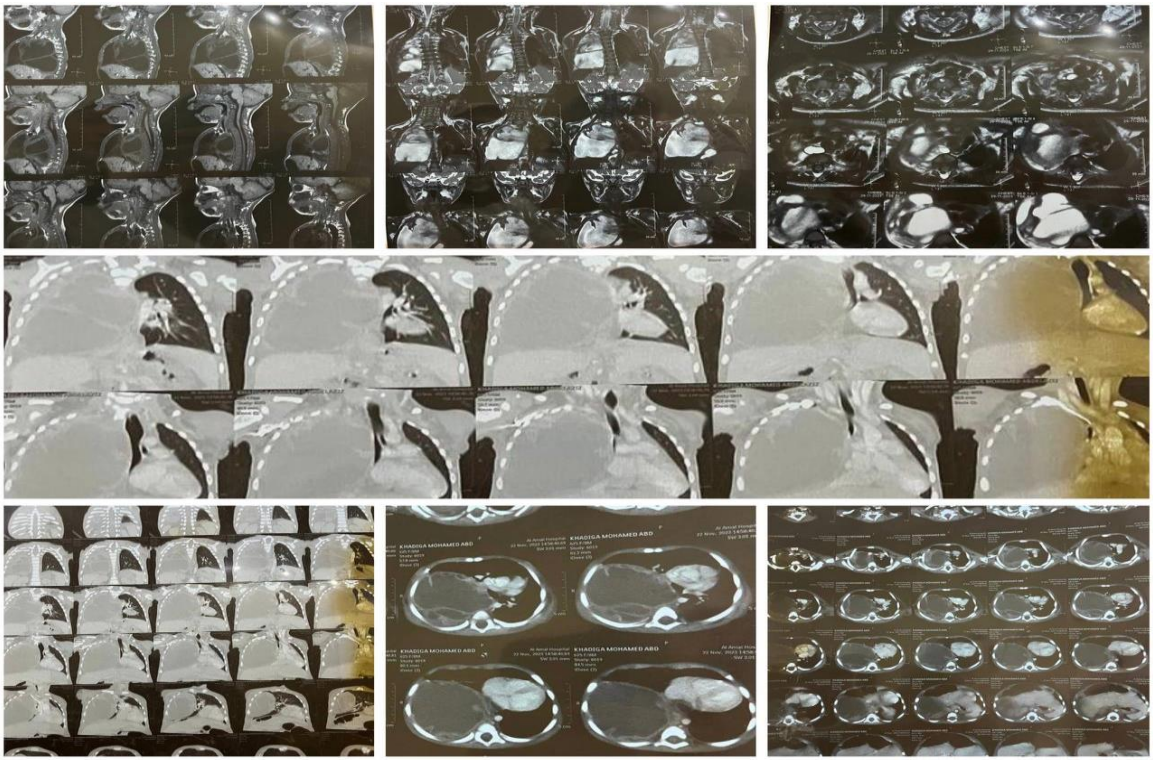
We report a case of a 5-month-old Female child, born at full term via normal delivery, weighing 3.980kg, , the patient was diagnosed antenatally at 36 weeks gestation via ultrasound, which revealed a cystic lung lesion. and the family history was notable for congenital heart disease, but the other siblings were healthy.

The child was apparently normal until 9 weeks of age when he developed a cough (dry type, with diurnal variation, aggravating by dust, chemical and, foreign body). There was a history of a fever, and fast breathing. On general physical examination, the child was FTT, anemic (hemoglobin –4.8g /dl) requiring two units of blood transfusion. The child was started on intravenous antibiotics but showed no improvement. significantly dropping HGB up to 3.6g/dl. She subsequently experienced acute gastroenteritis and was dehydrated, requiring further management and additional blood transfusion. Investigation for celiac disease and cow*s milk allergic returned normal. Bilateral crepitation was heard on chest auscultation. Blood and sputum cultures were negative. Chest X-ray was suggestive of right- sided pneumonia. Ultrasound thorax showed a large cystic lesion with turbid content. Contrast-enhanced computed tomography (CECT) chest showed.

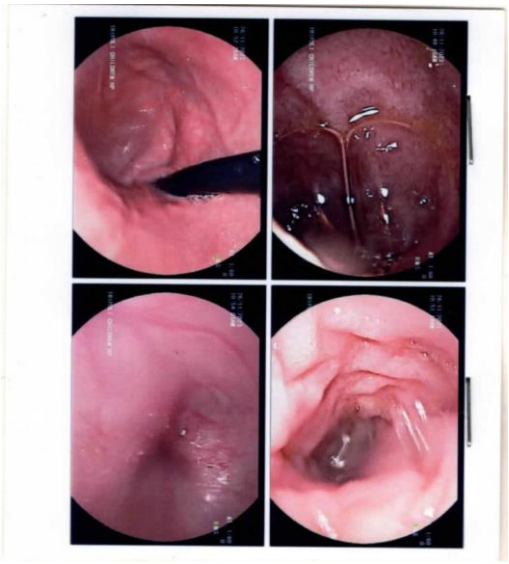


The imaging revealed a large right-lung cystic lesion with an enhanced wall, measuring approximately 8.4 × 7.4 × 8.0 cm. This collection is exerting a significant mass effect, resulting in compression of the remaining aerated right-lung tissue and a mediastinal shift toward the contralateral side. Additionally, multiple enlarged para-esophageal lesions were noted, most likely representing lymphadenopathy. The scan also demonstrated fusion of the D2 and D3 vertebral bodies..





Upper gastrointestinal endoscopy was performed and demonstrated normal findings, with no evidence of mucosal abnormalities or structural lesions.



Magnetic resonance imaging demonstrated a large lobulated, thick-walled cystic lesion measuring approximately 8 × 6 cm occupying most of the right hemithorax. The lesion exerts a marked mass effect, causing anterior and leftward displacement of the right lung as well as a contralateral mediastinal shift. No evidence of enteric communication was identified. A minimal right-sided pleural effusion was also noted.

Overall imaging features are most consistent with a congenital neurenteric cyst, possibly associated with a foregut duplication cyst. Additionally, there is multilevel vertebral segmentation abnormality, including fused C6–C7 vertebral bodies and a butterfly configuration of D1.

III) Management and surgical intervention

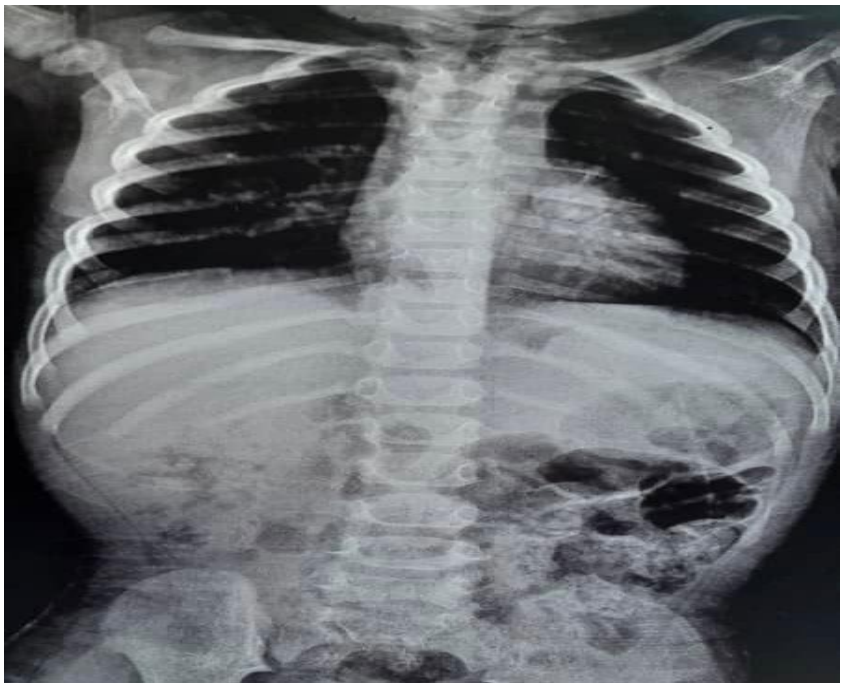
This retrospective longitudinal cohort study included 238 patients who presented with maxillofacial Due to persistent failure to thrive (FTT), the patient was initially managed with nasogastric tube (NGT) feeding, in combination with oral and intravenous proton pump inhibitors. At 9 months of age, her weight was recorded at 4.5 kg, indicating ongoing growth failure. Surgical intervention was initially postponed due to a concurrent chest infection but was eventually performed in May 2024, when the child was 14 months old.

During a right posterolateral thoracotomy, a cystic mass was identified in the posterior mediastinum, abutting the esophagus and extending caudally through a defect in the right hemidiaphragm into a cleft between the liver and right kidney. The cyst was easily separable from surrounding structures and contained clear, straw-colored fluid. At the time of surgery, the patient's weight had increased to 7 kg, reflecting significant progress in both growth and clinical status following the therapeutic interventions.

Histopathological examination revealed mild chronic gastritis in a gastric biopsy and a duodenal segment with reactive lymphoid aggregates. The cyst was classified as a Group B neurenteric cyst, containing complex elements of both the gastrointestinal tract and the tracheobronchial tree, including mucous glands and smooth muscle within its wall.

Postoperatively, the patient was admitted to the intensive care unit (ICU) for 24 hours and required a blood transfusion. Her C-reactive protein (CRP) remained elevated at 176.62 mg/L, and she received a two-week course of antibiotics.

At one-month follow-up, the patient demonstrated significant improvement in general condition and weight gain. Referral to a pediatric neurologist was made due to delayed motor development. By 19 months of age, her weight had increased to 9 kg, and she currently weighs 11 kg, reflecting continued catch-up growth and clinical improvement.



IV) Discussion

Enteric duplication cysts (EDCs) and neurenteric cysts are both rare congenital anomalies, which, although infrequent, can lead to significant clinical complications, including failure to thrive (FTT), anemia, and other systemic symptoms. EDCs, which may be associated with ectopic gastric mucosa in some cases, typically occur along the alimentary tract, with the ileum being the most common site. These cysts may cause clinical manifestations based on their size, location, and associated factors, such as the presence of gastric mucosa or other structural anomalies .Neurenteric cysts, on the other hand, arise from abnormal separation of the notochord and endoderm during embryogenesis and are often seen with other congenital abnormalities, particularly those involving the vertebrae, spinal cord, and gastrointestinal tract .Our case highlights a rare presentation involving both a neuroenteric cyst and other associated congenital anomalies, including vertebral abnormalities and failure to thrive.

The patient in this case was diagnosed with a congenital neuroenteric cyst, identified antenatally at 36 weeks via ultrasound, where a cystic lung lesion was detected. The clinical presentation of the child included persistent symptoms such as dry cough, fever, and fast breathing, which progressively worsened into significant failure to thrive (FTT) and anemia. At 9 weeks of age, the infant was noted to be anemic, with a hemoglobin level of 4.8 g/dL, which dropped further to 3.6 g/dL despite blood transfusion and antibiotic therapy. The development of acute gastroenteritis and dehydration necessitated further interventions. The presence of a large cystic lesion in the right lung and associated mediastinal shift, along with multiple vertebral anomalies, strongly suggested the diagnosis of a congenital neuroenteric cyst.

Neurenteric cysts often present with nonspecific symptoms depending on their size and location. They can cause respiratory symptoms due to their proximity to the lungs and other structures. In our case, the cyst was large (8.4 x 7.4 x 8.0 cm) and caused a significant mass effect on the surrounding lung tissue, resulting in pneumonia and a mediastinal shift. Imaging modalities such as contrast-enhanced computed tomography (CECT) and magnetic resonance imaging (MRI) were essential in delineating

the cyst's characteristics and confirming the diagnosis. The absence of enteric communication with the cyst further pointed to a neurenteric cyst, which is typically not directly linked to the gastrointestinal tract.

This patient also exhibited multiple vertebral anomalies, including D2 and D3 vertebral body fusion and abnormal segmentation of the cervical and thoracic vertebrae. Vertebral malformations are commonly associated with neurenteric cysts due to the abnormal embryologic development of the notochord and endoderm. The presence of these anomalies often complicates the clinical picture, as they may contribute to neurological and developmental delays, as seen in our patient who exhibited delayed motor development and required further neurologic evaluation. Additionally, the finding of mild chronic gastritis in the gastric biopsy emphasizes the potential for ectopic gastric mucosa within the cyst.

Management of neurenteric cysts usually involves surgical resection, especially in symptomatic cases, as these cysts can lead to a range of complications, including respiratory distress, gastrointestinal symptoms, and failure to thrive. In our case, surgery was postponed initially due to the child's ongoing chest infection but was eventually performed after the child showed partial recovery and reached a weight of 7 kg. The surgical approach included ht posterolrigateral thoracotomy and exploratory laparotomy.

Histopathological examination confirmed the diagnosis of a neuroenteric cyst, with features of more complex elements such as mucous glands and smooth muscle within the cyst wall, typical of Group B cysts. These cysts often contain elements of both the gastrointestinal tract and the tracheobronchial tree .Postoperatively, the child required intensive care for 24 hours and continued to receive antibiotics, as her inflammatory markers remained elevated. Blood transfusions were administered, and the patient gradually stabilized.

The patient showed significant improvement following surgery, with notable weight gain and a resolution of respiratory and gastrointestinal symptoms. At one year of age, the child had reached a weight of 9 kg, and currently, the patient weighs 11 kg. Although she made significant progress in her general condition, her delayed motor development warranted referral to a pediatric neurologist for further assessment and management.

V) Conclusions:

This case underscores the importance of early diagnosis and multidisciplinary management of congenital neuroenteric cysts in infants. Despite initial complications, timely surgical intervention and ongoing care led to significant improvement in the patient's health and weight. Further monitoring for developmental delays is recommended.

This case highlights the importance of early recognition and surgical intervention for congenital neuroenteric cysts, especially in the context of associated anomalies like vertebral malformations and enteric duplications. The clinical manifestations of these cysts can vary widely, and a multidisciplinary approach is often necessary for diagnosis and management. Prompt surgical resection can lead to significant improvement in symptoms, as evidenced by the patient's postoperative progress. However, ongoing developmental monitoring and supportive care are essential for addressing long-term outcomes. The association of such cysts with other congenital malformations emphasizes the need for comprehensive evaluation and care in affected children.

VI) Consent

Informed written consent for the publication of this case report and any accompanying images was obtained from the patient’s parents prior to submission.

VII) ACKNOWLEDGEMENTS

We would like to extend our heartfelt gratitude to the patient's family for their unwavering support and cooperation throughout the course of treatment. Their dedication and involvement played a significant role in the patient's recovery and overall well-being. We appreciate their trust and collaboration, which contributed to the success of the care provided."

VIII) References:

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(6) Bhattacharya A, Samujh R, Rao KL, Mittal BR. Long segment jejuno-ileal duplication cyst with ectopic gastric mucosa detected on 99mTc-pertechnetate scintigraphy. Indian J Nucl Med. 2013; 28:96–8. [PMC free article] [PubMed] [Google Scholar] Given the complexity of the cyst and its location, careful dissection was required to avoid injury to surrounding structures, including the lung and mediastinum.