Case Report

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A Unique Case of Duplication Cyst of Esophagus and Ileum, with Meckle's Diverticulum

Fatima Majeed[1]*, Prod Muhammad Saleem[2], Imran Hashim[3], Ayesha Manzoor[3] and Nisha Shan[4]

[1]*FCPS Pediatric Surgeon, King Edward Medical University/Mayo Hospital, Pakistan*

[2]*The Children Hospital and University of Child Health Sciences CHUCH Lahore, Pakistan*

[3]*CHUCH, Pakistan*

[4]*King Edward Medical University/ Mayo Hospital, Lahore, Pakistan*

***Corresponding author:** Fatima Majeed, FCPS Pediatric Surgeon, King Edward Medical University/Mayo Hospital, Lahore, Pakistan

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Introduction

Enteric duplication cysts is a rare congenital anomaly, that can be present anywhere along gastrointestinal tract, from mouth to anus [1]. Mostly its treatment is surgical resection [2]. Here we present a case of duplication cysts of esophagus and ileum with Meckel's diverticulum, which is a rare entity. There is only one case previously reported that shows enteric duplication cyst with Meckel's diverticulum.

Case Presentation

A 6months old female baby, with history of recurrent episodes of chest infections and respiratory distress since 20th days of life, was referred to Pediatrics Surgery Department, The Children's Hospital and the Institute of Child's Health, Lahore. Patient had history of melena and hematemesis for which she was transfused with packed cells multiple times. Patient was admitted in medical ICU and had persistently deteriorating saturation and blood gasses, for which she was intubated. Meckel's bleed scan was suggestive of active bleeding site in small intestine. CT angiogram showed benign posterior mediastinal cystic lesion/infected collection. CT chest with contrast showed multiple vertebral fusion anomalies in upper thoracic spine with bilateral bifid 1st rib and thick-walled cystic lesion with suspicion of neuroenteric, bronchogenic or duplication cyst. Endoscopy was done that showed normal esophageal, gastric and duodenal mucosa and histopathology report showed mild duodenitis. Patient was operated in emergency on 10-7-19 with suspicion of ruptured thoracic cyst as patient had clinical signs of shock. Right thoracotomy with cyst excision was done. Intraoperatively, 4*2cm cyst was seen in right paravertebral thoracic region extending from posterior mediastinum up to left of vertebral column. Biopsy report came out to be esophageal duplication cyst. Later on, patient had good recovery but her melena and hematemesis didn't settle. On 28-7-19, she was operated for abdominal symptoms and exploratory laparotomy with excision of small gut duplication and end to end ileo-ileal anastomosis was done. Patient had small gut duplication that was starting 2feet from deudeno-jejunal junction up to distal ileum, 20cm proximal to ICJ. Patient had uneventful postoperative recovery. Later on biopsy report showed duplication cyst with Meckel's diverticulum (Figures 1-8).

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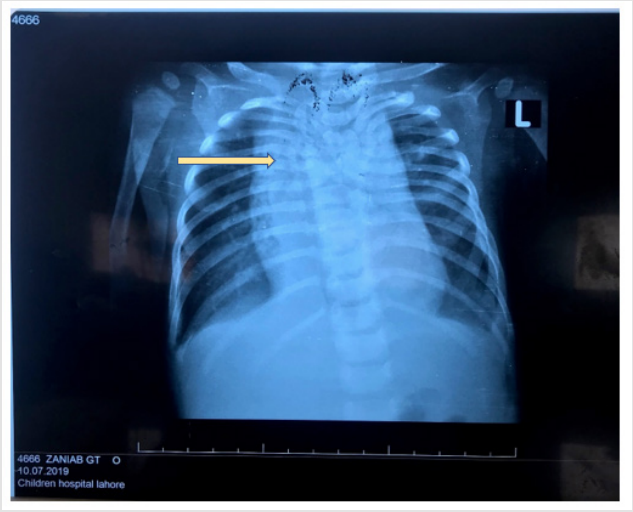

Figure 1: Showing chest x ray postero-anterior view. Arrow shows widened mediastinum.

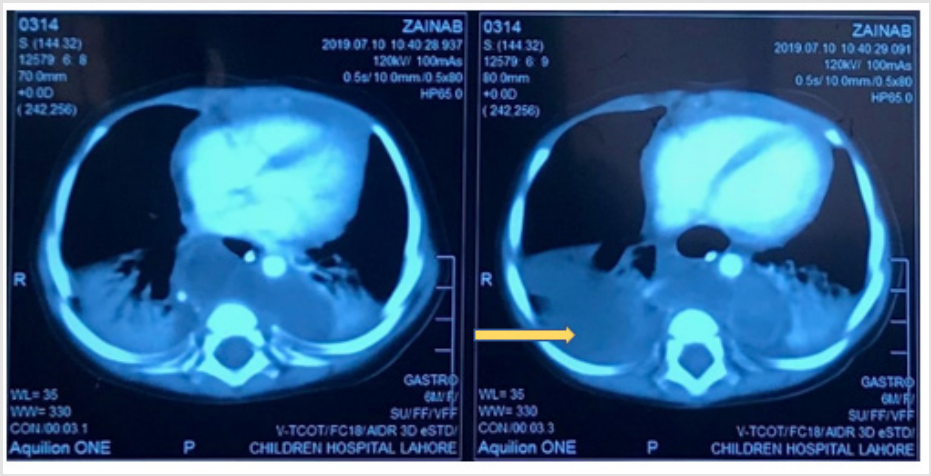

Figure 2: CT chest, Arrow showing cyst in posterior mediastinum.

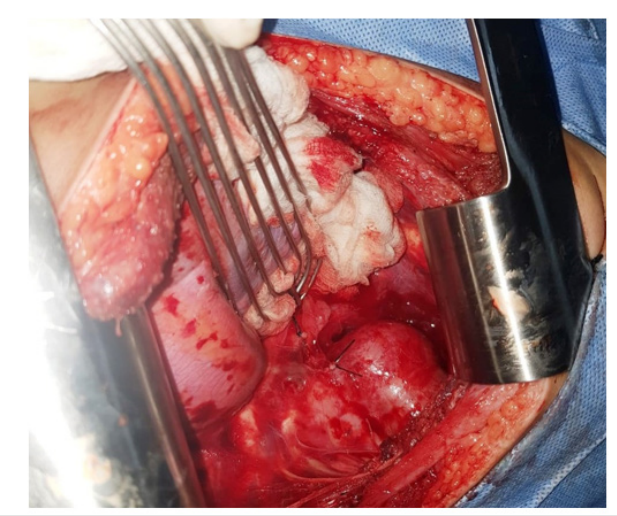

Figure 3: Arrow showing mediastinal duplication cyst..

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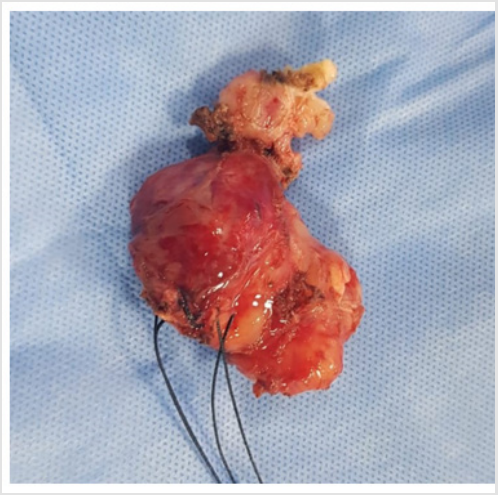

Figure 4: Showing excised duplication cyst of mediastinum with silk sutures for marking.

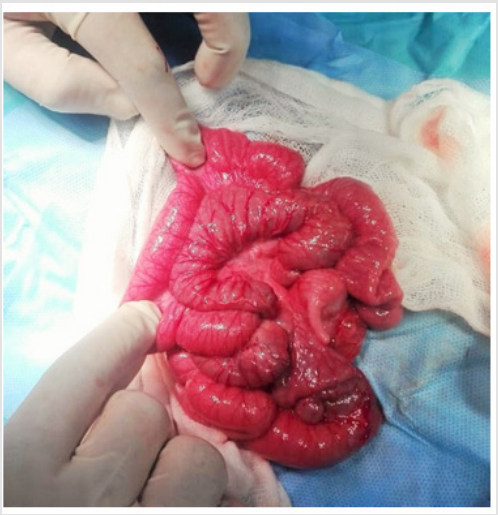

Figure 5: Showing enteric duplication.

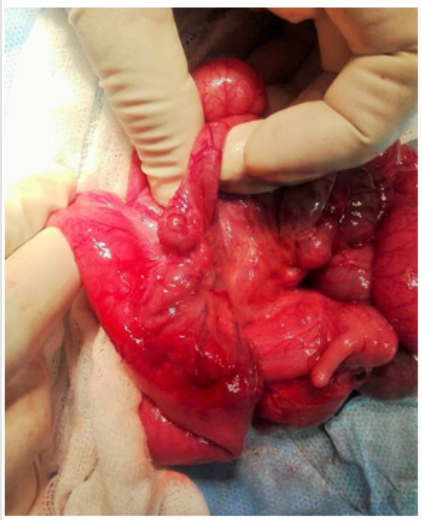

Figure 6: Arrow showing terminal end of duplication of ileum.

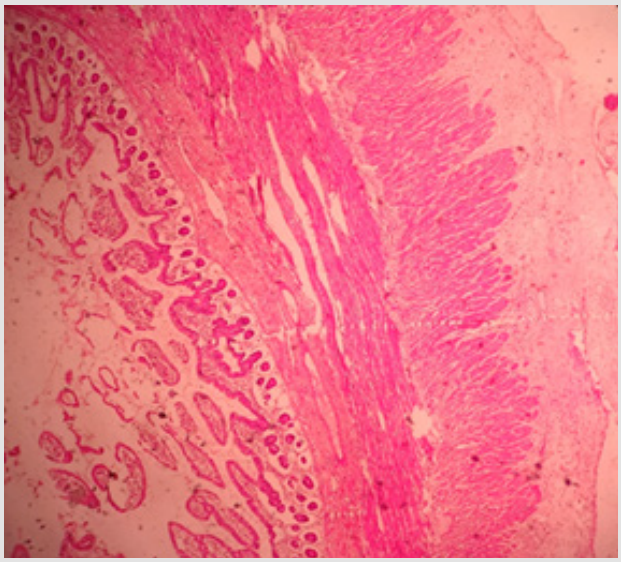

Figure 7: Thoracic cyst Mucosa with muscle coat.

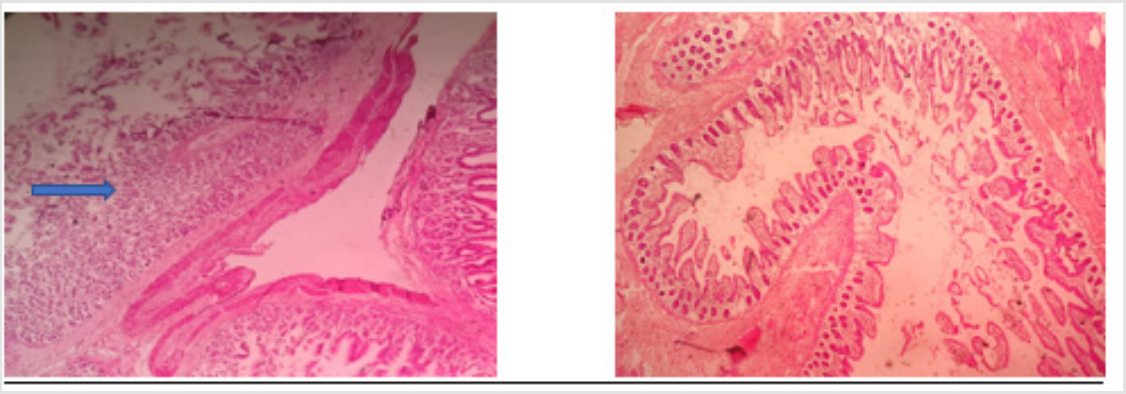

Figure 8: Enteric Duplication Cyst with Meckel's diverticulum. Arrow showing ectopic gastric mucosa.

Discussion

Enteric duplication cysts usually present in neonatal life and early infancy, with more than 80% presenting before 2 years of age [3]. All of them must have three components:

1. Hollow structure,
2. Lined with gastrointestinal tract epithelium, and
3. Have a wall of smooth muscle often connected to intestinal wall [1,4].

They are mostly located on mesenteric border and more commonly found in ileum than esophagus [2]. Mostly they have same blood supply as of adjacent intestine [5]. The etiology is unknown and many theories have been presented to explain the development of enteric duplication cysts which includes aberrant luminal recanalization theory, abortive twinning theory, split notochord theory, diverticular theory, and environmental factors [1,6] The symptoms and clinical picture are variable and nonspecific, with most of them depending upon site, size, presence of ectopic mucosa, inflammation and communication with bowel. Within abdomen, they may present with pain, nausea, vomiting, intestinal obstruction, perforation, intussusception, intestinal bleeding or may be identified as an incidental finding intra operatively [1,3]. In thorax they may be asymptomatic or they may present with respiratory distress, cough, pain, progressive dysphagia and recurrent pneumonias [7,8]. Diagnosis is made with the help of ultrasound, barium studies or CT scan. Ultrasound is investigation of choice for intraabdominal masses and tells about nature of mass. CT scan shows relation of mass with surrounding structures. Barium studies in upper gastrointestinal tract shows filling defects. Endoscopy and radioisotope scanning can detect site of bleeding. MRI and endoscopic ultrasound can also be used. However, all these cannot confirm diagnosis of duplication cyst, that is only possible after resection and histopathological examination. [1] As Meckel's diverticulum has its own separate blood supply and

is present on antimesenteric border, so it is easily resected. But, in case of duplication cyst, due to common blood supply, it is treated by excision and anastomosis [3].

In present case, Meckel's diverticulum was present along with duplication cyst, that is a rare entity. GI bleed scan showed bleeding site in small intestine but could not localize EDC in thoracic cavity. Whereas, Kumar.K et.al, showed that, GI bleed scan has 75% sensitivity in detecting EDC with ectopic gastric mucosal tissue [9]. Differential diagnosis of enteric duplication cyst was made but it was confirmed after surgery. We have very few case reports showing multiple duplication cysts in chest and abdomen simultaneously. Moreover, only one case is reported for EDC with Meckel's diverticulum [10,11].

Conclusion

Enteric duplication cysts are rare entity and one should look for multiple cysts in chest and abdomen if single one is found. Moreover, presence of Meckel's diverticulum with EDC should be kept in differential diagnosis.

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