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Continuous Thoraco-Abdominal Duplication in Children Type Ia: Case Report

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Abstract **Case Report**

Introduction: Thoraco-abdominal duplications, rare malformations of the oesophagus gastrointestinal tract, present diagnostic challenges often requiring surgical confirmation. ***Observation:*** A 22-month-old male with a thoraco-abdominal mass. After laparotomy, histological analysis confirmed an esophagogastroduodenal duplication extending from the esophagus to the duodenum. ***Discussion:*** The esophagogastroduodenal duplication extending from the esophagus to the duodenum is rare, with variable presentation and embryological origins. Clinical manifestations vary, imaging aids diagnosis, and surgery is the definitive treatment. ***Conclusion:*** This case underscores the importance of prompt identification and management of digestive duplications, emphasizing the necessity of resection and potential adjacent tissue involvement.

Keywords: Thoraco-abdominal duplication, continuous thoraco-abdominal duplication, esophagogastroduodenal duplication, surgical excision, case report.

INTRODUCTION

Thoraco-abdominal duplications are defined as tubular or cystic malformations located in a segment of the digestive tract, from the oral cavity to the anus. They represent a rare entity, with an incidence of 1:4500, as reported by Schalamon *et al*., [1]. This condition was first described by Ladd in 1937 [2] and accounts for only 2% of cases of gastrointestinal duplications, according to Thamri *et al*., [3]. Antenatal diagnosis is possible in cases of large cystic forms [4].

Cystic or tubular, these duplications are always situated on the mesenteric border of the gastrointestinal tract, sharing a common blood supply. However, the etiology remains poorly characterized [5]. One explanation involves abnormalities in the development of the neurenteric canal, often associated with anterior or posterior spina bifida, as described by Bentley and Smith in their "split notochord syndrome" theory [6].

Other proposed etiological theories include disruptions in the recanalization process of the embryonic gut or the persistence of embryonic diverticula, which may explain short-segment duplications without additional anomalies [7]. Clinical manifestations vary depending on the site of the duplication, with the majority of cases presenting within the first year of life. Although the diagnosis can sometimes be made preoperatively through imaging or other investigations, these malformations are frequently discovered during surgery [8].

CASE OBSERVATION

A 22-month-old male infant, born from a monitored pregnancy with normal antenatal ultrasounds, presented with a history of poorly tolerated iron deficiency anemia requiring two transfusions. At one year of age, he developed intermittent abdominal distension and dyspnea, with no associated malformations.

Clinical examination revealed a conscious infant with mild conjunctival pallor, stable hemodynamics, abdominal distension without tenderness, and no hernial defects. Abdominal ultrasound identified two cystic formations adjacent to the kidneys, suggesting digestive or renal origin. Abdominal CT showed a large retroperitoneal cystic mass (108 mm), while thoraco-abdominal MRI confirmed a well-defined, biloculated thoraco-

abdominal duplication extending from the esophagus to the duodenum, with digestive wall features.

Surgical laparotomy revealed a type Ia esophagogastroduodenal duplication along the mesenteric border, with separate blood supply. Histological analysis revealed gastric tissue with well-differentiated glandular structures and inflammatory changes, confirming its compatibility with esophagogastroduodenal duplication. Postoperative recovery was uneventful.

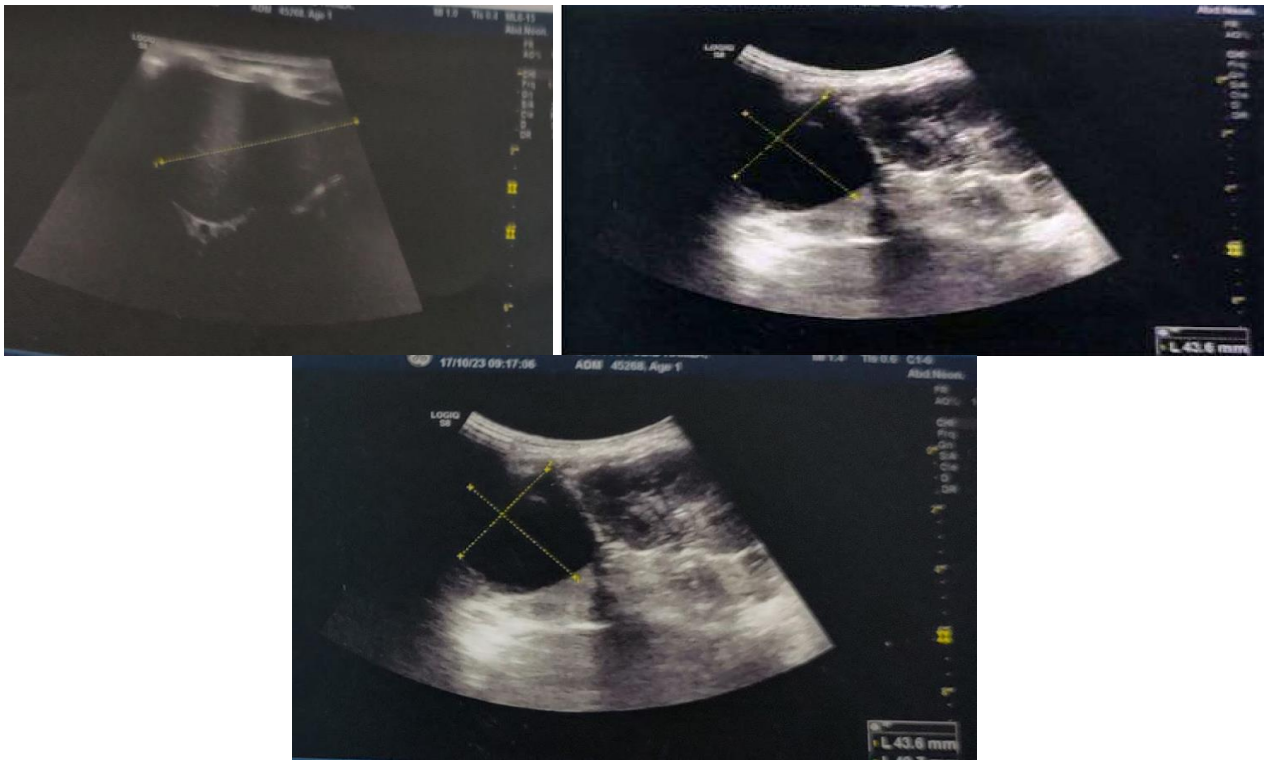

Figure 1: Abdominal ultrasound revealed two liquid formations

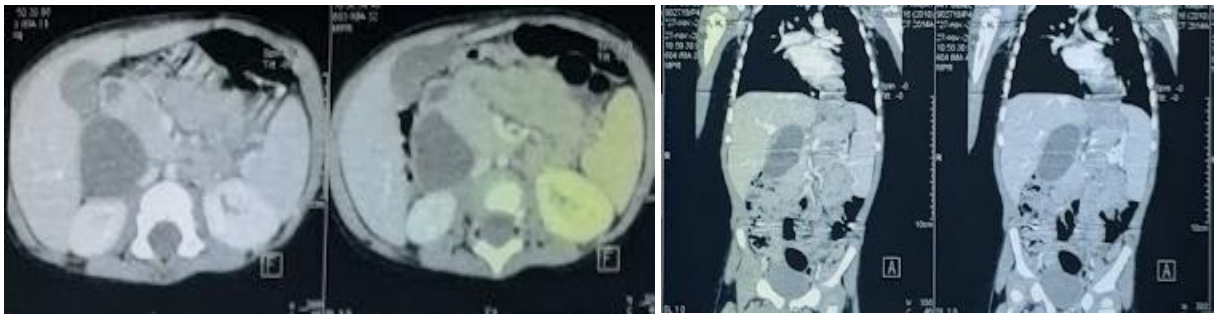

Figure 2: Abdominal CT scan revealed a sizable cystic mass with a clean wall occupying the bilateral retroperitoneal region

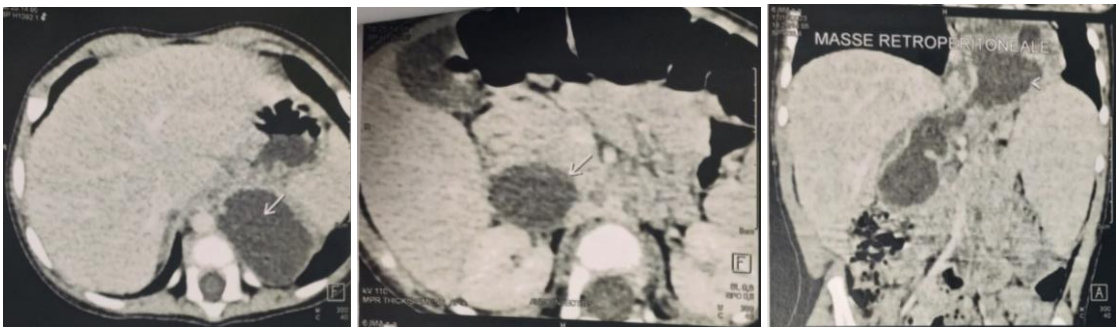

Figure 3: Thoraco-abdominal MRI showed a supra and subdiaphragmatic mass evoking a thraco-abdominal duplication: esophagogastroduodenal duplication extending from the esophagus to the duodenum

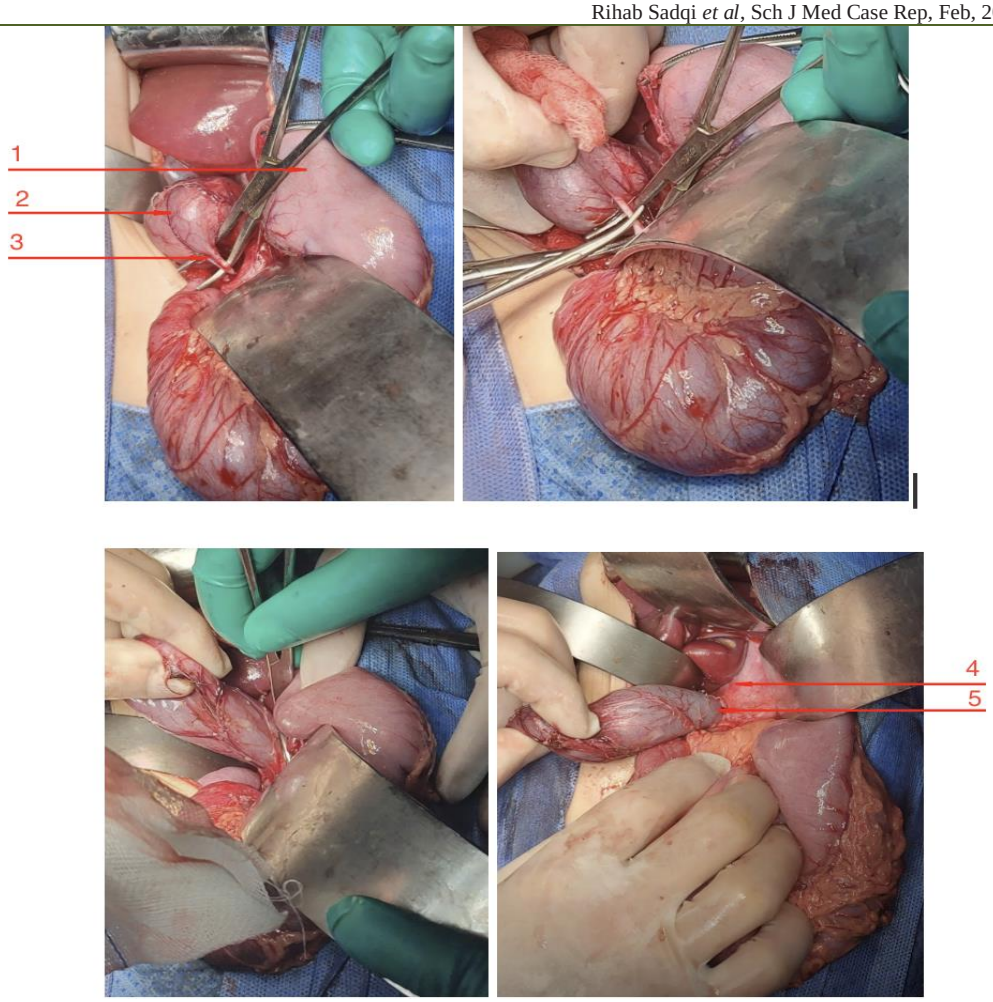

Figure 4: Intraoperative imaging of an esophagogastroduodenal duplication demonstrating continuity and communication between the esophagus and the duodenum. 1: stomach, 2: duplication zone, 3: communication with the duodenum, 4: diaphragm, 5: esophagogastroduodenal duplication

DISCUSSION

Thoraco-abdominal duplications are rare congenital malformations, accounting for only 2% of gastrointestinal duplications, with possible communication with the digestive tube in 60% of cases. The first case was described by Calder in 1733 and later popularized by Ladd in 1937 [2]. These anomalies are often present in early childhood, with 70% of cases diagnosed before the age of two, though some may remain asymptomatic into adulthood [9].

The antenatal discovery is increasingly being described [3]. Elsewhere, they are usually diagnosed during the first year of life in ¾ of cases by respiratory signs due to compression of the airways by chest mass or digestive signs following secretion by the atopic mucosa [3, 10, 11].

The etiology remains uncertain, with proposed theories including the split notochord syndrome, partial twinning, persistent embryological diverticula, or aberrant luminal recanalization [12]. Vascular classification divides duplications into two types: Type I, where the duplication lies on one side of the mesentery with parallel arterial supply, and Type II, where it is located between mesenteric leaves, with arteries traversing both surfaces of the duplication. Rare vascular anomalies, such as the absence of the inferior vena cava, have also been reported in association with thoraco-abdominal intestinal duplications [13, 14].

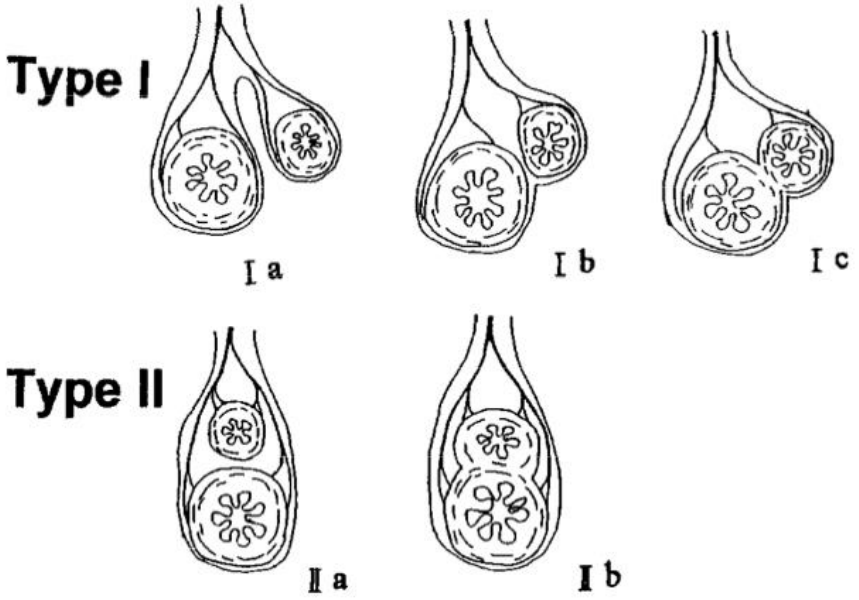

Figure 6: Vascular Classification for Small Intestinal Duplications [14]:

- Type I: parallel type: - Type Ia: duplication with a separated mesentery, -Type Ib: sharing the common mesentery with bowel, -Type Ic: sharing the common muscular coat with the bowel
- Type II: intramesenteric type: - Type IIa: separating from the normal bowel, -Type IIb: sharing the common muscular coat.

Clinical presentation varies by location and size, ranging from abdominal pain, vomiting, and obstruction to asymptomatic masses [13, 15]. Ultrasound is the primary diagnostic tool, often revealing cystic intra-abdominal lesions. While CT and MRI can provide additional details, especially for thoraco-abdominal or duodenal duplications, they may not add significant value for small intestinal duplications [1, 16]. In some cases, barium studies have proven diagnostic, particularly when gastric emptying issues are suspected [17].

Surgical resection remains the standard treatment, often involving complete excision of the duplication with adjacent bowel [17]. Histopathological examination is crucial for confirming the diagnosis, as it reveals the characteristic features of ectopic mucosa and other tissue anomalies.

Although ileal duplications are the most common (over 60%), thoraco-abdominal and colonic duplications are rare, with the latter accounting for only 6.8% of cases [15]. The variability in clinical presentation is influenced by factors such as ectopic mucosa, inflammation, or communication with adjacent structures [6].

According to a case series, 17 patients exhibited a connection between the duplication and the normal gastrointestinal tract. Among these, 16 connections were identified below the diaphragm, with only one case involving the cervical esophagus (Table I) [18]. While the type of mucosal lining appeared to influence patient symptoms, no clear association was found between the type of lining and the location of the communication. Gastric mucosa was identified in six duplications, and respiratory epithelium was observed in one. In our case, we identified gastric mucosa and observed communication between the esophagogastroduodenal duplication and the duodenum. The duplication was continuous along the mesenteric border, with a separate blood supply. As reported by McLetchie *et al*., the duplication presented as a mass on the right side of the hypochondrium [19].

Table I: Site of communication of the reported thoracoabdominal duplications with normal intestinal tract [18]

Site of communication	No.
Jejunum	9
Duodenum - First portion - Third portion	 -3 -1
Ileum	2
Cervical oesophagus	1
Not known	1
None	8

If possible, the entire duplication should be removed during one operation. This is to avoid leaving a closed, noncommunicating sac containing acid-secreting gastric mucosa [18]. As in our case, the entire esophagogastroduodenal duplication was resected.

Early diagnosis facilitates elective surgery under optimal conditions, improving outcomes. The role of advanced imaging and histopathology remains pivotal in the comprehensive evaluation and management of these rare anomalies.

CONCLUSION

The case reports highlight the challenge of diagnosing through imaging and confirming with surgical and pathological examination. Minimally invasive diagnostic and therapeutic approaches are preferable. Early identification and treatment of this congenital anomaly are crucial, with mandatory resection of the duplication and potentially adjacent normal intestine.

Competing Interests: The authors declare no competing interest.

Author Contributions:

All authors contributed to the creation of this article. The authors also declare that they have read and confirmed the final version of this article.

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