The Ileoileal Intussusception due to a Tubular Duplication in a Child: A Case Report

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Abstract

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Title page

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Abstract
Intussusception is a surgical emergency. If not immediately treated, it can lead to bowel wall perforation. In 2.2-15% of cases, they have pathologic lead points such as Intestinal duplication. This case report presents a rare tubular ileal duplication as a necrotic Ileoileal intussusception in a 4-year-old girl. **Key Clinical Message** In all children over three years old with intussusception suspicious to pathologic lead points and initially negative ultrasonography (US) results never exclude second repeat US. **Keywords**

Intussusception, Ileal duplication, Alimentary tract duplication

1 INTRODUCTION
Intussusception is one of the most frequent intestinal obstruction etiologies when a portion of the gastrointestinal tract gets telescoped into the near bowel segment. The most common type is the idiopathic ileocolic one (98%). This can happen in children ages six months and two years. in the past, it had high mortality and morbidity rates which, with the progress of diagnosis and effective treatment, came to a good outcome. In cases not immediately treated, bowel wall ischemia and perforation are probable, with an unfavorable
prognosis. The classic symptoms that increase the clinical suspicion of it are a triad of vomiting, palpable abdominal mass, and passage of "currant jelly stools" (stools mixed with blood and mucus).

In 2.2-15% of cases, they have pathologic lead points (PLPs) such as:

- Meckel diverticulum (the most common lead point)
- Intestinal duplication (relatively rare)
- Benign polyps
- Malignant lymphoma
- Peutz–Jeghers syndrome
- and hamartoma (n = 1).

In non-complicated intussusception, radiological reduction is the treatment of choice, and the failure rate of it is 10-60 %. In these cases, surgical therapy is mandatory. Intestinal duplication is a rare congenital malformation (one in 10000 birth) and is defined as alimentary pathway duplication, mostly involving the midgut, especially the ileum. Duplication by itself has many types. One of them is the tubular type, which may be short or involve entire segments, and it is rare. We herein report a rare case of a tubular ileal duplication as a complicated Ileoileal intussusception in a 4-year-old girl.

2 Case presentation

2.1 History

A four-year-old Afghan refugee girl without medical or surgical history was admitted to the pediatric ward with the chief complaint of acute abdominal pain.

The symptoms started as intermittent abdominal pain (every 60 minutes with a duration of 20 minutes) in the periumbilical and hypogastric areas three days ago, accompanied by two times bilious vomiting (last night and one hour before admission) and one time of currant jelly stool defecation with no sign of hematochezia. Her complaints were temporarily relieved by taking anticholinergics and antiemetics as self-treatment.

2.2 Physical Examination

In general appearance, she was an ill child, and physical examination revealed an abdominal distention with a tender palpable mass in the periumbilical area.

2.3 Laboratory data and Imaging study

Laboratory results showed a leukocytosis (white blood cell (WBC):15600/MCL), neutrophilia (absolute count of polymorphonuclear cells: 12948/MCL), C-reactive protein level (4.4 mg/dL), and ketonuria (acetone:4+) (WBC:4-6/hpf, epithelial cell: 0-1/hpf, few bacteria) in urine analysis.

Ultrasonography (US) confirmed an ileal–ileal invagination as a "doughnut" sign with obstructive findings, free abdominopelvic fluid, and no vascular flow in invaginated Loup in doppler investigations.

2.4 Treatment plan

Due to the exhibiting symptoms of Peritonitis, we candidate her for urgent operation after fluid resuscitation and administration of pre-operative broad-spectrum antibiotics. Under general anesthesia, the stomach was decompressed with a nasogastric tube. Exploratory laparotomy showed a twisted Ileoileal invaginated part at 20 cm from the ileocecal valve. (Figure 1a) At first, we tried to untwist the volvulus, then freed the 20 cm invaginated part by milking from the distal region. Eventually, the 10 cm necrotic area was approved as the small intestine duplication (Figure 1b). Because of the whole thickness necrosis and the impossibility of recovering the invaginated part's circulation, the decision was made to accomplish resection and anastomosis surgery. The patient was discharged from the hospital after 96 hours without any complications or specific events. Histopathological findings confirmed pan-necrosis in the resected specimen. (Figure2)

3 Discussion
Intussusceptions are usually idiopathic, and just in 2.2-15% of cases, they have pathologic lead points such as Meckel diverticulum (most common) and Intestinal duplication.

Most PLPs present as ileoileocolic intussusceptions (40% of all ileoileocolic), generally, signs and symptoms are similar. Still, presentations can vary and are often nonspecific because of the wide range of lesions or intestinal anomalies. PLPs should be considered, especially in the case of multiple recurrences and failure of enema technique reduction or children over five years.

We recommended in suspicious circumstances; a primarily negative US investigation should never exclude a second repeat US.

Enteric duplication is often in the ileum and may be either cystic or tubular. Unlike the Meckel diverticula, most are attached to the mesenteric portion and consist of two parts: an outer layer of smooth muscle and an inner lining of the gastrointestinal epithelium.

It should be embedded that many children may represent non-classic symptoms, so diagnostic evaluation depends on radiologic imaging to make the precise diagnosis. In the hands of a skilled examiner, ultrasound (US) is the choice for confirming or excluding intussusception and duplication in children with 98-100% sensitivity and 88-100% specificity. However, the computed tomography (CT) scan is the gold standard in adults, while in the US is more common.

Treatment of a stable patient with intussusception begins with an enema reduction in the emergency department; in the absence of contraindications such as perforation or shock signs, etc.

And the presented case came with the classic symptoms of intussusception that US findings boosted the suspicion. At the operation, the tubular type of ileal duplication, which is almost rare, was seen, and ileoileal intussusception was approved.

We also searched extensively four databases of PubMed, Cochrane Library, Scopus, and Google Scholar with MeSH terms of the keywords of intussusception, duplication, ileal duplication, tubular duplication, and children with the 5-year time filter, English language filter, and no article type filters and for exclusion criteria, we omitted irrelevant studies. We found few cases of children with intussusception secondary to tubular ileal duplication.

4 Conclusion
The authors recommend that surgeons consider pathologic lead points, even though there are routine imaging and sonographic and laboratory findings, especially in children over five years with failed reduction ones.

AUTHOR CONTRIBUTIONS
Hojjat Shaman Zahroodi wrote original draft, Paria Dehghanian and Mehran Monazzami was involved in data interpretation, editing, and review. Khashayar Atqiace was engaged in manuscript preparation, reporting and planning and review.

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CONFLICT OF INTEREST
The authors declare that they have no competing interests in this original work.
DATA AVAILABILITY STATEMENT

The datasets are available from the corresponding author upon reasonable request.

ETHICAL APPROVAL

We confirm that all named authors have read and approved the manuscript. The protection of the intellectual property associated with this manuscript has been our consideration.

CONSENT

The authors confirmed that they had gotten all proper patient written consent formats. The parents have given their consent for their images and other clinical information to be reported in the form. The parents were informed about the confidentiality of the names and initials, and efforts would be made to hide their identity.

References

Figures' legends:

Figure 1a: Twisted Invaginated Intestinal Part

Figure 1b: The 10 cm necrotic part appeared as the duplication of the Small Intestine

Figure 2: Histopathological findings of the duplicated segment of

a: Small intestinal wall with hyperplastic lymphoid follicles (H&E staining, low-power field).

b: Pan-mural hemorrhagic infarct of the intestinal wall with lymphoid follicles remnants