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# Case Report on Ileoilleal Knotting with Multiple Congenital Adhesion and Enteroentric Fistula in A 19-Year-Old Female Ethiopian

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**Abbreviations:**

SBO: Small Bowel Obstruction; CT: Computed Tomography

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## 1. Abstract

### 1.1. Introduction and Importance

Ileoileal knotting is an exceptionally rare cause of small bowel obstruction characterized by the formation of a true knot between two segments of the ileum, resulting in closed-loop obstruction and potential bowel ischemia. The presence of multiple congenital adhesions with entero-enteric fistulas further complicates the diagnosis and surgical management. This case highlights the diagnostic challenges and importance of early surgical intervention of this rare case presentation.

### 1.2. Case Presentation

We report a 19-year-old female with a 17-year history of recurrent episodes of abdominal pain, who presented with acute exacerbation of epigastric pain and vomiting of 20 hours duration. Intraoperative findings revealed a 540° counter-clockwise volvulus with ileoileal knotting, extensive Grade III and IV adhesions, two enteroenteric fistulas, and ischemic bowel loops. Surgical management involved exploratory laparotomy, de-rotation, adhesiolysis, division and repair of fistulas, and bowel preservation. Histopathology showed reactive lymphoid hyperplasia without malignancy, granuloma, or necrosis.

### 1.3. Clinical Discussion

Ileoileal knotting is rarely reported, and its coexistence with multiple congenital adhesions and enteroenteric fistulas makes this case unique. The diagnostic challenge stems from nonspecific presentations that mimic other acute abdominal conditions. Prompt surgical intervention remains the cornerstone of management to minimize morbidity and mortality.

### 1.4. Conclusion

This case underscores the importance of early recognition and surgical management of ileoileal knotting complicated by congenital adhesions and fistulas. It contributes to the limited literature on this rare condition and highlights the need for heightened clinical suspicion in atypical presentations of intestinal obstruction.

## 2. Introduction and Importance

Ileoileal knotting is an exceptionally rare cause of small bowel obstruction characterized by the formation of a true knot between two segments of the ileum, resulting in closed-loop obstruction and potential bowel ischemia. The presence of multiple congenital adhesions further complicates diagnosis and surgical management, as these fibrous bands may contribute to bowel fixation and obstruction. This case report details the presentation, diagnosis, and surgical treatment of a 19-year-old female with a 17-year history of recurrent abdominal pain who presented acutely with epigastric pain and vomiting, later found intraoperatively to have a 540° counter-clockwise volvulus with an ileoileal knot complicated by extensive Grade III and IV adhesions, two sites of enteroenteric fistula, and ischemic bowel loops. Surgical intervention involved exploratory laparotomy, de-rotation, adhesion release, fistula division and repair, and bowel preservation. Histopathological examination revealed reactive lymphoid hyperplasia with no evidence of malignancy, granuloma, or necrosis, supporting a benign inflammatory response rather than

infectious or neoplastic pathology. Postoperative recovery was uneventful with favorable outcomes at six months follow-up. This report underscores the diagnostic challenges posed by this rare condition and highlights the importance of early surgical intervention to reduce morbidity and mortality. It contributes to the limited literature on ileoileal knotting compounded by multiple congenital adhesions and enteroenteric fistulas, emphasizing the need for heightened clinical suspicion in atypical presentations of intestinal obstruction.

## 3. Introduction and Importance

Ileoileal knotting is an exceedingly rare cause of intestinal obstruction [1-6[ It refers to knot formation between two ileal segments [7]. It is the obstruction of an intestinal segment with closed loop phenomenon secondary to knotting of the mesentery [4]. Congenital adhesions are also rare findings in adults [8]. Ileum is the commonest site of congenital bands, followed by colon, mesentery, omentum, peritoneum, jejunum as well as every site of the gastrointestinal tract, including the abdominal organs and peritoneum [9].

While there are reports of ileoileal knotting are rare with fewer than two dozen documented cases globally [2,8], specific cases that detail the combination of this condition with multiple congenital adhesions are even scarcer. The presence of congenital adhesions can complicate the clinical picture by anchoring bowel segments and creating fixed points conducive to volvulus and knot formation, making diagnosis and surgical intervention more challenging [8,9]. These fibrous bands are often asymptomatic but can cause mechanical obstruction or facilitate complex bowel pathologies when multiple and extensive. Notably, the ileum is the most common site for congenital bands, followed by the colon and mesentery [9]. The diagnostic challenge is compounded by the nonspecific clinical presentation, often mimicking other causes of acute abdomen and typically requiring intraoperative confirmation [7,10].

This case report presents a uniquely complex scenario of a 19-year-old female with ileoileal knotting, compounded by multiple congenital adhesions and two enteroenteric fistulas-an extremely rare constellation. The patient's history of recurrent epigastric pain without prior abdominal surgery, along with intraoperative findings of extensive adhesions and ischemic bowel, emphasizes the diagnostic challenges and underscores the importance of early surgical intervention. This case aims to contribute to the limited literature on this condition and highlight the need for heightened clinical suspicion in similar atypical presentations.

## 4. Case Presentation

This is a 19-year-old female patient who presented with burning epigastric pain that radiates to the back. The pain is aggravated by sitting up and lying back in recumbent position, and is relieved by leaning forward. The pain has lasted for 20 hours. Additionally, she experienced five episodes of non-bilious, non-projectile vomiting of ingested material over the same duration. The patient has a history of a milder similar complaints occurring every year since she was two years old, with symptoms that resolve spontaneously, and she didn't seek medical attention prior to her current presentation. She has no history of failure to pass feces or flatus, and no other pertinent medical history.

Upon admission, the patient appeared acutely ill, with a pulse rate between 124 and 128 beats per minute. Her abdomen was flat and moved with respiration. There were no visible peristaltic movements, but there was direct tenderness in the right upper quadrant and the epigastric area. A digital rectal examination revealed a collapsing rectum with minimal hard stool.

An abdominal ultrasound showed twisting of the small bowel mesentery and superior mesenteric vessels, indicating an inverted relationship between the superior mesenteric artery and the superior mesenteric vein, with a clockwise whirlpool sign on Doppler study. Mild dilation of the proximal small bowel loops was also observed. Additionally, there was a well-defined left adnexal cystic lesion measuring 5 cm by 5.5 cm, which had an anechoic center and posterior acoustic enhancement, with no solid components detected.


**Figure 1**: This is abdominopelvic X-ray reveals a dilated bowel loop, which is suggestive of intestinal obstruction. The dilated loop, visible predominantly on the left side of the abdomen, exhibits prominent gas patterns, consistent with air-fluid levels or accumulation of gas due to stasis. This radiographic finding correlates well with the intraoperative diagnosis of ileoileal knotting, where the entanglement of intestinal loops leads to mechanical obstruction and subsequent bowel dilation.

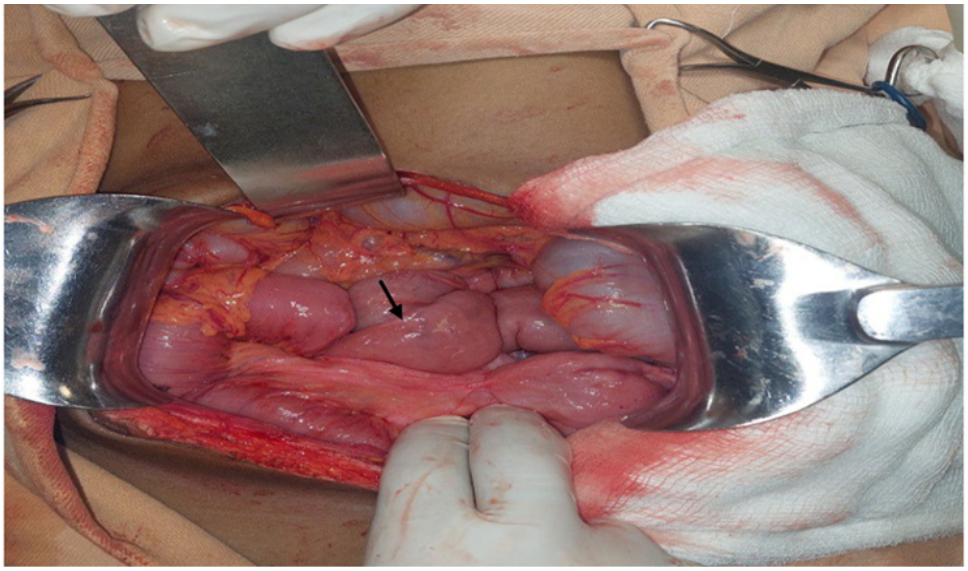

**Figure 2**: This image shows that the black arrow highlights the exact site of the ileoileal knotting, where two segments of the ileum have twisted tightly around each other, leading to obstruction and compromised blood flow one of the critical causes of the patient's condition.

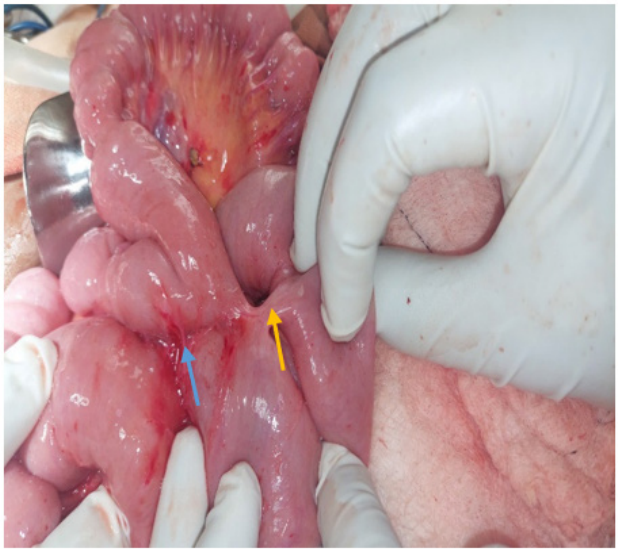

**Figure 3**: This illustration shows key pathological findings: the black arrow indicates the site of knotting, which suggests a potential obstruction. The blue arrow points to congenital adhesions, the green arrow highlights mesenteric lymphadenopathy, and the red arrow marks a distended ileal loop that is ischemic, indicating compromised blood flow and potential tissue damage in that segment of the intestine.

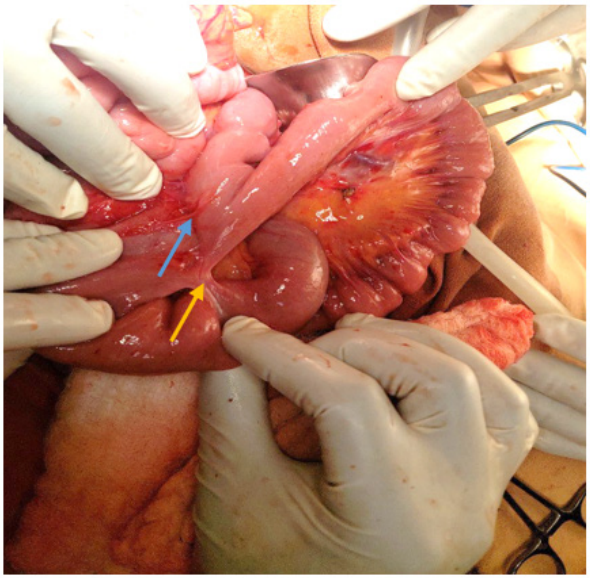

**Figure 4 and Figure 5:** This surgical image, clearly illustrates key pathological features. The yellow arrow points to a fistulous tract, likely resulting from chronic inflammation or pressure changes, while the blue arrow highlights dense congenital adhesions, which may have contributed to the bowel entanglement and obstruction seen in this case.

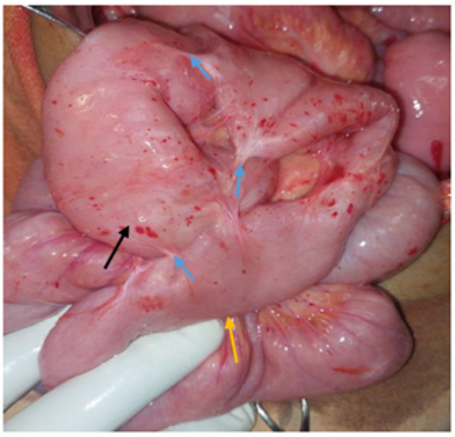

**Figure 6**: This photograph, highlights several significant findings. The blue arrow indicates multiple dense adhesions, likely contributing to the bowel entrapment and obstruction. The black arrow points to a dilated ileal loop, suggestive of upstream accumulation due to the obstructive process. The yellow arrow identifies a fistulous connection, possibly formed as a result of prolonged inflammation or pressure-related injury.

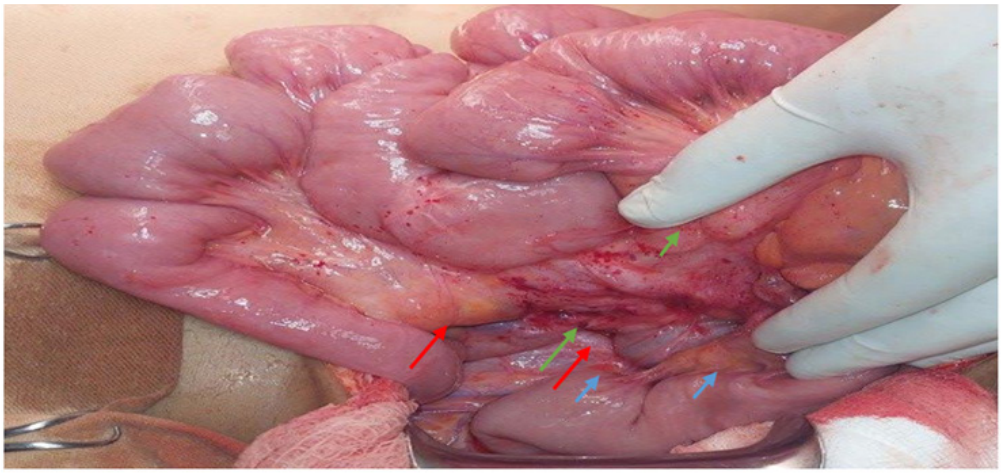

**Figure 7**: The intraoperative image, shows several key pathological findings: the black arrow highlights the area of ileoileal knotting, the red arrow points to a distended ischemic ileal loop, the blue arrow indicates congenital adhesions, and the green arrow identifies mesenteric lymphadenopathy. These findings correlate with the underlying pathology observed during the procedure.

## 5. Differential Diagnosis

### 5.1. Small Bowel Volvulus

Twisting of a loop of small intestine causing obstruction and ischemia.

Can cause similar whirlpool sign on imaging and acute abdomen presentation.

### 5.2. Adhesive Small Bowel Obstruction

Due to post-surgical or congenital adhesions causing mechanical obstruction.

Common cause of SBO and can present with intermittent or acute symptoms.

### 5.3. Internal Hernia

Herniation of bowel through a defect in the mesentery or peritoneum.

Presents with obstruction symptoms and can be complicated by strangulation.

### 5.4. Intussusception

Telescoping of one bowel segment into another causing obstruction.

More common in children but possible in adults; can cause ischemia and acute pain.

### 5.5. Ileocecal or Ileosigmoid Knotting

Other types of intestinal knotting aside from ileoileal knotting, e.g., ileosigmoid knot.

Presents similarly with acute obstruction and ischemic bowel.

### 5.6. Mesenteric Ischemia

Acute interruption of blood flow to the intestines causing ischemic pain and obstruction-like symptoms.

Can be due to embolism, thrombosis, or volvulus.

### 5.7. Acute Pancreatitis

Epigastric pain radiating to the back, nausea/vomiting.

Needs to be differentiated as it has overlapping symptoms but different management.

### 5.8. Gastrointestinal Perforation with Peritonitis

Could present with acute abdomen and signs of obstruction secondary to paralytic ileus.

Ovarian Cyst Complications (e.g., Torsion or Rupture)

Given the adnexal cyst found on imaging, gynecological causes can mimic acute abdomen and vomiting.

### 5.9. Gastric Outlet Obstruction

Due to peptic ulcer disease, malignancy, or other causes leading to vomiting and epigastric pain.

Surgical Intervention;-Intraoperative finding; There was Counter-clockwise rotation (5400) of the small bowel, with 20 cm of the terminal ileum entangled at the volvulus base with multiple firm lymphadenopathies, thickening the small bowel mesentery. There was also Extensive Grade III and IV adhesions with ischemic loops causing various area of complete obstruction. Three diverticula found at the distal 20 cm of the terminal ileum and an enteroenteric fistula at 2 sites.

Likely left ovarian cyst measuring 10 cm by 6 cm was also found with Cecum fixed in the right lower quadrant with the duodeno-jejunal flexure positioned at L1.

Exploratory laparotomy + De-rotation + Adhesion release was done.

Where the volvulated small bowel was carefully de-rotated, and an enlarged segment of the small bowel was found to be fixed. Obstructive adhesions, classified as Grade IX, were identified and released, including two sites of enter-enteric fistulas which were divided and repaired transversely. Biopsies were taken from several areas of mesenteric lymphadenopathy and sent for histopathological examination. A thorough peritoneal lavage was performed using warm normal saline. After confirming the correct surgical count, the abdomen was closed meticulously in layers.

### 5.10. Postoperative Course

The patient tolerated the procedure well and was on post-operative care and follow up. She was discharged after a weeks of

hospital stay. She has continued to be in good health after six months of follow-up.

Pathology Result;-(Mesenteric lymph nodes) Reactive lymphoid hyperplasia (mixed follicular hyperplasia with sinus histiocytosis). Microscopic sections reveal three encapsulated tissues that exhibit relatively preserved lymphoid architecture, featuring variably sized secondary follicles and significant sinus dilatation.

No granulomas or necrosis are observed, and no malignant cells are present.

## 6. Clinical Discussion

Intestinal obstruction remains one of the leading surgical emergencies worldwide, and the subset small bowel obstruction Small bowel obstruction (SBO) remains a common and urgent surgical challenge globally, accounting for up to 80% of intestinal obstructions, particularly in resource-limited settings like Ethiopia, where adhesions, volvulus, and hernias dominate the etiological spectrum [1]. Among these causes, intestinal knotting,particularly ileoileal knotting,is exceedingly rare, with very few cases reported in the literature ]2,5]. The coexistence with multiple congenital adhesions and fistulas markedly increases its rarity and complexity.

Intestinal knotting First described nearly 500 years ago by Riverius in the 16th century, intestinal knotting remains an uncommon cause of mechanical bowel obstruction [5]. Bowel knots can be classified into two types: true knots, in which one intestinal segment passes through a loop formed by another intestinal segment, and pseudo knots, in which the intestinal segments are wrapped around each other without actual formation of a knot [7].

True ileoileal knots occur when one ileal loop slips through a pre-existing loop and forms a closed-loop obstruction. This mechanical entrapment often leads to rapid vascular compromise and bowel ischemia [7]. The addition of multiple congenital adhesions may contribute to anchoring of bowel loops, increasing the risk of volvulus and knotting by providing fixed points around which the mobile bowel can twist [8].

What makes this case exceptionally unique is the presence of two enteroenteric fistulas, an extremely uncommon finding in conjunction with ileoileal knotting. Fistula formation in such settings is usually secondary to chronic low-grade inflammation, ischemia, or prolonged pressure necrosis, all of which can be promoted by adhesive bands and obstructive pathology [10]. The formation of these fistulas in a young patient without any previous abdominal surgery strongly suggests a congenital or developmental anomaly as the underlying contributor [8,9] further complicating the clinical course and surgical decision-making.

Diagnosis of ileoileal knotting is done intraoperatively [1,5,10] Most reported cases are confirmed only upon surgical visualization of the knot [5,7]. Patients usually present acutely with abdominal distension and pain. While plain abdominal X-ray may demonstrate obstruction, ultrasound findings such as the ‘whirlpool sign’ indicating mesenteric vessel twisting as seen in our case were pivotal in prompting urgent laparotomy. Computed tomography may provide additional detail when indicated [10].

The etiology of intestinal knotting including ileo-ileal knotting is unknown [1,2,4]. It occurs more frequently in regions with high rates of small bowel and sigmoid volvulus, potentially linked to high-fiber diets and ileal hypermotility [1,2,4]. Anatomical factors like a relatively long small bowel mesentery or a capacious abdomen are implicated [2].

Unlike typical presentations, this patient’s recurrent, self-resolving symptoms and absence of prior abdominal surgery highlight an atypical clinical course that challenges conventional diagnostic pathways. Critically, our case demonstrates how congenital adhesions serve as pivot points facilitating the 540° volvulus and knot formation [7,9], while chronic adhesive pathology led to enteroenteric fistulas, likely chronic sequelae of recurrent micro-obstructions [9] These fistulas further anchored bowel segments, exacerbating volvulus and ischemia through rigid fixation points.

The discovery of two enteroenteric fistulas intraoperatively adds significant pathophysiological insight. These fistulas typically arise from chronic inflammation, pressure necrosis, or untreated perforations [10]. In this context, they likely developed over years due to recurrent subclinical obstructions from congenital adhesions, creating aberrant connections between ileal segments. Their presence underscores the chronicity of the patient’s condition despite episodic symptoms, and they may have exacerbated obstruction by forming rigid fixation points that potentiated volvulus [7].

This triad (knotting, adhesions, fistulas) represents a unique pathomechanism scarcely documented in literature. They also necessitated meticulous adhesiolysis, de-knotting, and primary repair. These findings reinforce the complex interplay of congenital factors and mechanical forces in generating rare but life-threatening pathologies. This case adds significant value to the limited literature and highlights the need to consider congenital adhesions and fistulous complications in young patients with unexplained recurrent abdominal symptoms.

The condition of mesenteric lymph nodes in this patient initially raised suspicion due to their firmness and prominence during surgery. However, histopathological analysis revealed reactive lymphoid hyperplasia with mixed follicular hyperplasia and sinus histiocytosis, without evidence of granuloma, necrosis, or malignancy. This indicates a benign reactive process, possibly secondary to chronic inflammatory or obstructive stimuli, rather than an underlying neoplastic or granulomatous disease.

Such lymphoid hyperplasia likely reflects a local immune response to prolonged low-grade inflammation associated with chronic adhesive pathology, recurrent ischemia, or the presence of fistulous tracts. In young patients with congenital abnormalities, this pattern is not uncommon and must be interpreted in the context of clinical and intraoperative findings.

### 6.1. Treatment Strategies

Delay in diagnosis will increase the morbidity and mortality. One study that evaluated the management of small bowel obstruction reported mortality of up to 9.5%, and 34.9% of patients had complications at presentation [1] The mortality rate can go as high as about 50% [4].

For complex presentations involving fistulas:

• Resuscitation with IV fluids, nasogastric decompression, and broad-spectrum antibiotics remains urgent [1,4].

• Intraoperatively, fistulas demand meticulous management:

o Division with wide resection of fibrotic tracts

o Transverse repair to minimize stenosis risk [4]

o Assessment of distal patency to exclude secondary obstructions

Bowel viability dictates resection decisions: When the bowel loops are viable, careful untying of the loops is performed as there is very low risk of recurrence. Non-viable segments require resection with exteriorization/anastomosis (1,4,5,10). When fistulas coexist, resection must include involved segments to prevent recurrence [3].

There is no consensus as yet as to whether the ileo-ileal knot should be released or not before resecting the bowel. Some suggest that the knot should be released first to determine the extent of salvageable small bowel, so as to avoid excessive bowel resection [7]

In our case, although ischemic segments were observed, they were salvageable following detorsion and adhesiolysis. The two enteroenteric fistulas were successfully divided and closed primarily, which is the recommended approach when bowel viability is preserved and there is no gross contamination [10].

Given the benign nature of mesenteric lymphadenopathy (reactive lymphoid hyperplasia), no further oncologic or infectious workup was warranted. Pathology confirmed the reactive nature of lymph node enlargement, likely secondary to chronic inflammation from the adhesions and fistulas. This underscores the importance of routine lymph node biopsy in atypical intraoperative findings to exclude more serious etiologies and guide postoperative management appropriately.

Postoperatively, the patient should be monitored for hydration status, anemia, and signs of anastomotic leak (heightened risk in fistulous bowel). Follow-up for signs of short bowel syndrome (given terminal ileum involvement) should be instituted depending on the length of resected gut and symptoms exhibited by the patients [4].

## 7. Conclusion

This case report documents an exceptionally rare combination of ileoileal knotting, multiple congenital adhesions, and dual enteroenteric fistulas in a young female with no history of prior surgery-an occurrence not previously well-documented in the literature. It highlights how chronic adhesive pathology can drive acute surgical emergencies through fistulization and mechanical fixation. The chronicity of her symptoms and intraoperative findings suggest a congenital origin that led to gradual but significant anatomical and physiological disruption.

The unique triad challenges conventional diagnostic pathways, emphasizing the need for:

• Preoperative vigilance for ‘whirlpool sign’ on ultrasound

• Intraoperative inspection for fistulas in congenital adhesion cases

• Aggressive resection of fistulous segments to prevent recurrence

Its educational value lies in demonstrating how congenital anomalies may underlie recurrent abdominal symptoms in young adults, necessitating early surgery to avert catastrophic outcomes.

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