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## Chilaiditi syndrome in pediatric patients - Symptomatic hepatodiaphragmatic interposition of colon: A case report and review of literature

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### Abstract

#### BACKGROUND

Chilaiditi syndrome is a rare disorder characterized by the hepatodiaphragmatic interposition of the intestine.

#### CASE SUMMARY

Here we report a case of a 12-year-old male who was admitted to the pediatric intensive care unit secondary to abdominal pain and severe respiratory distress. He was treated conservatively but the symptoms persisted requiring a surgical approach. While there have been several cases of Chilaiditi syndrome reported in adults, there is a scarcity of cases reported in the pediatric population. Our review of the literature found only 30 pediatric cases, including our reported case, with Chilaiditi syndrome, 19 (63%) of which were male. The median age of diagnosis was 4.5 years old with an interquartile range of 2.0-10.0 years. In our review, we found that the most common predisposing factors in children are aerophagia (12/30 cases) and constipation (13/30 cases). Ninety percent of the cases

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presented with complete intestinal interposition, in 100% of which, the colon was involved. Three of the 30 cases were associated with volvulus.

#### CONCLUSION

In the pediatric population, conservative (21/30 cases) and surgical (8/30 cases) treatment approaches have produced satisfactory outcomes for all the patients, regardless of approach.

**Key Words:** Abdominal pain; Dyspnea; Constipation; Rare diseases; Respiratory insufficiency; Colon; Case report

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**Core Tip:** We describe a pediatric case of Chilaiditi syndrome with successful treatment, as well as a literature review of all pediatric case reports of Chilaiditi syndrome. In the pediatric patients, both conservative and surgical approaches in treating Chilaiditi syndrome with treatment of predisposing factors have resulted in satisfactory outcomes.

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## INTRODUCTION

Chilaiditi syndrome, first described by Viennese radiologist Dr. Chilaiditi[1] in 1910, is noted to be an extremely rare disorder associated with various symptoms including nausea, vomiting, abdominal pain, constipation, and respiratory distress. The condition is recognized radiologically by the presence of the hepatodiaphragmatic interposition of the intestine, called Chilaiditi sign. Chilaiditi sign can be confused radiologically with other conditions such as pneumoperitoneum and subdiaphragmatic abscess. The cause of Chilaiditi syndrome is currently unknown, but may include intestinal, diaphragmatic, or hepatic factors. While most cases can be managed conservatively, a few cases require surgical intervention[2]. We report a pediatric case of Chilaiditi syndrome and a literature review of a pediatric case series of Chilaiditi syndrome.

## CASE PRESENTATION

### Chief complaints

A 12-year-old male was admitted to the pediatric intensive care unit due to severe respiratory distress.

### History of present illness

With this present admission, the patient presented with respiratory distress and right upper quadrant abdominal pain. He was placed on oxygen supplementation *via* nasal cannula to maintain normal oxygen saturations.

### History of past illness

Prior to this admission, he experienced persistent cough, dyspnea, nausea, and chest pain for over two months. He was prescribed antibiotics, nebulizations, and pain medication; however, there were no improvements in his respiratory symptoms. The patient has a history of asthma, gastroesophageal reflux disease, constipation, and a prior diagnosis of Chilaiditi syndrome. The diagnosis of Chilaiditi syndrome was made two years prior to this admission when the patient presented with a one-week

history of right upper quadrant pain, nausea, and vomiting. There was no history of recent weight loss. An abdominal computerized tomography (CT) showed constipation and colonic interposition between the liver and the diaphragm with displacement of the liver (Figure 1). Constipation was initially managed with a routine bowel cleansing protocol and a daily stool softener; however, intermittent episodes of abdominal pain persisted.

#### **Personal and family history**

No relevant family history.

#### **Physical examination**

No relevant physical examination.

#### **Laboratory examinations**

Laboratory results from complete blood count, comprehensive metabolic panel, and C-reactive protein were within normal limits.

#### **Imaging examinations**

A chest X-ray revealed that the transverse colon was above the liver. On the first hospital admission day, a kidney, ureter, and bladder X-ray (KUB) showed significant amount of fecal material and air-filled colonic loops which were slightly dilated and reaching the right hemidiaphragm (Figure 1).

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## **FINAL DIAGNOSIS**

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A final diagnosis of Chilaiditi syndrome was given.

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## **TREATMENT**

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He subsequently received a bowel-cleaning regimen with GoLytely®. A follow-up KUB on the second hospital admission day showed the resolution of fecal retention or constipation. However, the patient continued to complain of tachypnea and right upper quadrant pain. Because of his persistent respiratory and abdominal symptoms, and due to the lack of significant improvement, surgery was consulted. The patient underwent laparoscopic colopexy and peritoneal abrasion of the diaphragm and liver. Significant intraoperative findings included a redundant transverse colon, no evidence of volvulus or adhesions in the upper abdomen, a relatively small right liver lobe (noncirrhotic), and a large gap between the liver and the anterior chest wall and diaphragm.

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## **OUTCOME AND FOLLOW-UP**

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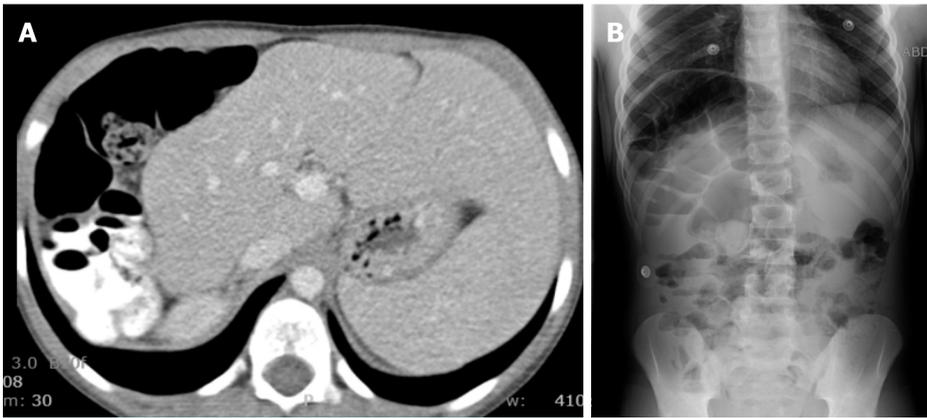
His respiratory distress and abdominal pain resolved completely post-operatively and the patient was discharged with a maintenance stool softener regimen, colonic stimulant, and adequate dietary fiber. At the one-month follow-up after surgery, the patient reported regular bowel movements and no recurrence of his respiratory distress. He reported some mild intermittent episodes of right upper quadrant abdominal pain but never required emergency care or any interventions since the surgery.

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## **DISCUSSION**

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The essential hallmark of Chilaiditi sign in Chilaiditi syndrome is that the air-filled loops of intestine remain unchanged in position of the patients due to its immobilization in a relatively limited space between the liver and the anterior chest wall[3]. Chilaiditi sign may be described as an incidental finding on plain radiological studies in asymptomatic patients. It is thought to occur in 0.025% to 0.28% of the general population. It is markedly more prevalent in the elderly and in men. This increased prevalence in the elderly suggests that it is an acquired rather than a congenital



**Figure 1** Imaging of abdomen and pelvis of a 12-year-old male with Chilaiditi syndrome and constipation. A: Computerize tomography. Marked air and fecal retention of the entire colon with colonic interposition above the liver with displacement of the liver leftwardly. This phenomenon indicates segmental agenesis of the right lobe of the liver and relaxation of the hepatic suspensory ligament; B: Plain X-ray. Markedly greater than average amount of fecal material particularly in the rectosigmoid colon. Few air fluid levels in the distal small bowel and air filled colonic loops that reach the right hemidiaphragm.

condition. Torgersen reported the prevalence of Chilaiditi syndrome to be 0.2% in men older than 65 years and 0.02% in men 15-65 years, with a male to female ratio of 4:1[4]. Murphy *et al*[5] associated Chilaiditi syndrome with being overweight or obese. Five of his ten patients found to have Chilaiditi syndrome on abdominal CT were obese (850 patients in the study, 10 of whom had Chilaiditi syndrome)[5]. In obese patients, a significant amount of fat accumulates between liver and diaphragm, with secondary widening of potential space, which is subject to substantial swings in pressure during the respiratory cycle. Following the same concept, the increased proportion of intraabdominal fat among men compared with women might explain the increased prevalence of Chilaiditi syndrome in men[6]. While there have been severe cases of Chilaiditi syndrome reported in adults, there is a scarcity of cases reported in the pediatric population. Our review of the literature found only 30 pediatric cases with Chilaiditi syndrome, 19 (63%) of which were male (Table 1). The median age of diagnosis was 4.5 years old with an interquartile range of 2.0-10.0 years[7-28].

The etiology of Chilaiditi syndrome has been categorized into (1) Intestinal: megacolon, abnormal colonic motility or redundancy, constipation, and congenital malrotation; (2) Hepatic: cirrhosis, segmental agenesis of the right lobe of the liver, and relaxation of the hepatic suspensory ligament; and (3) Diaphragmatic: phrenic nerve injury and diaphragmatic eventration[15,17]. Several risk and predisposing factors have been associated with this entity including, aerophagia, adhesions, obesity, constipation, mental retardation, pregnancy, muscular dystrophy, and significant weight loss[17,22]. Very rarely, episodes of volvulus have been associated to this syndrome, especially in the elderly population and could be complicated with cecal perforation[4,7,22,29,30]. Chilaiditi syndrome can further be divided in two types, depending on the degree of intestinal interposition and liver displacement: (1) In the complete form, the colon typically lies above the liver, there being contact between the liver and diaphragm, with the liver displaced inferiorly, anteriorly, and medially; and (2) In the incomplete (partial) form, the colon does not typically rise above the liver, but lays lateral or posterior to it[23]. In theory, patients after orthotic liver transplantation will have some degrees of intestinal interposition with the transplanted liver being displaced inferiorly, anteriorly, and medially.

In our review of the pediatric literature, we found the most common predisposing factors in children to be aerophagia (12/30 cases) and constipation (13/30 cases). Ninety percent of the cases presented with complete intestinal interposition, in 100% of which the colon was involved. Three of the 30 cases were associated with volvulus. In the case we described here, the predisposing factor was believed to be a combination of constipation, redundant colon, and intestinal dysmotility, associated with a relatively small right lobe of the liver, in turn, allowing a big space between the liver and the anterior chest wall and diaphragm.

The most common clinical presentation of Chilaiditi syndrome is constipation, abdominal pain, nausea, vomiting, abdominal distention, and respiratory distress. On physical examination, it is possible to encounter loss of hepatic dullness on percussion (Joubert sign)[7,8,23,25]. The diagnosis of hepatodiaphragmatic interposition can be demonstrated with radiologic tests such as a plain KUB, a right upper quadrant

Table 1 Case series of Chilaiditi syndrome in the pediatric population

Ref.	Sex	Age	Predisposition	Bowel segment	Symptoms	Type of interposition	Dx procedure	Treatment	Type of surgery	Outcome
[24]	M	16 mo	Aerophagia	Colon	Abdominal distention and pain, vomiting	Complete	KUB	Conservative	---	Resolution
[24]	F	3 yr	Aerophagia	Colon	Abdominal distention and pain, passed flatus	Complete	KUB	Conservative	---	Resolution
[24]	F	5 yr	Aerophagia, constipation	Colon	Abdominal pain, constipation	Partial	KUB	Conservative	---	Resolution
[24]	F	4 yr	Aerophagia	Colon	Anorexia, recurrent abdominal pain, vomiting	Complete	KUB	Conservative	---	Resolution
[27]	F	2 yr	---	Colon	Marasmus, vomiting, lethargy, inability to walk	Complete	KUB	Surgery (Volvulus)	Laparoscopic colopexy and transverse colectomy	Resolution
[26]	M	6 mo	---	Colon	Abdominal pain, vomiting	Partial	KUB	Conservative	---	Resolution
[19]	M	8 yr	Aerophagia	Colon	Abdominal pain, distention	Complete	KUB	Conservative	---	Resolution
[7]	M	12 yr	---	Colon	Respiratory distress, pleuritic pain, fever	Complete	CXR, BE	Surgical (Volvulus)	Laparoscopic detorsion	Resolution
[22]	M	17 yr	Mental retardation, constipation, congenital adhesions	Colon	Abdominal distention, vomiting, constipation	Complete	CXR, KUB	Surgical (Volvulus)	Laparoscopic transverse colectomy	Resolution
[28]	F	9 yr	Constipation	Colon	Abdominal pain, nausea, constipation	Complete	CXR, KUB	Conservative	---	Resolution
[9]	F	11 yr	Constipation	Colon	Abdominal pain, vomiting	Complete	KUB, CT	Surgery	Laparoscopic transverse colectomy	Resolution
[23]	F	9 yr	DE, constipation	Colon	Epigastric pain, constipation, nausea	Complete	CXR, KUB, CT	Conservative	---	Resolution
[23]	M	1 yr	DE	Colon	Recurrent respiratory distress	Complete	CXR, KUB, CT	Surgery	Correction of diaphragmatic eventration and elevation of right hemidiaphragm	Resolution
[23]	F	16 mo	DE	Colon	Recurrent respiratory distress	Complete	CXR, KUB, CT	Surgery	Correction of diaphragmatic eventration and elevation of right hemidiaphragm	Resolution
[25]	M	5 mo	---	Colon	Recurrent respiratory distress	Complete	CXR, CT	Conservative	---	Resolution
Present study	M	12 yr	Constipation	Colon	Recurrent respiratory distress, abdominal pain, constipation	Complete	CXR, KUB	Surgical	Laparoscopic colopexy	Resolution
[33]	F	2 yr	Constipation	Colon	Recurrent respiratory distress, abdominal pain,	Complete	CXR	Conservative	---	Resolution

					constipation						
[10]	M	8 yr	Constipation	Colon	Abdominal pain, constipation	Complete	KUB, CT	Conservative	---		Resolution
[11]	M	10 d	Constipation	Colon	Abdominal distension, respiratory distress, constipation	Complete	KUB	Conservative	---		Resolution
[12]	F	3 yr	Constipation	Colon	Recurrent respiratory distress, constipation	Complete	CXR	Conservative	---		---
[13]	M	4 yr	Aerophagia	Colon	Respiratory distress	Complete	CXR	Conservative	---		Resolution
[14]	M	6 yr	---	Colon	Abdominal pain, emesis, FTT	Complete	CXR	Surgical	Laparoscopic colopexy		Resolution
[15]	M	10 yr	Aerophagia	Colon	Recurrent respiratory distress	Complete	CXR, MRI	Conservative	---		Resolution
[15]	M	7 yr	Aerophagia	Colon	Recurrent respiratory distress, abdominal distention	Complete	CXR, MRI	Conservative	---		Resolution
[8]	M	4 yr	Aerophagia, constipation	Colon	Recurrent respiratory distress, abdominal pain, constipation	Complete	CXR, CT	Conservative	---		Resolution
[16]	M	3 yr	Aerophagia	Colon	Recurrent respiratory distress, abdominal distention	Complete	CXR	Conservative	---		Resolution
[17]	M	20 yr	Duchenne muscular	Colon	Recurrent respiratory distress	Complete	CT	Conservative	---		Resolution
[18]	M	19 yr	Dystrophy, aerophagia, constipation	Colon	Chest pain, respiratory distress, abdominal pain	Complete	CXR	Conservative	---		Resolution
[20]	F	1 yr	Aerophagia	Colon	Respiratory distress	Partial	CXR	Conservative	---		Resolution
[21]	M	10 yr	Constipation, mental retardation	Colon	Respiratory distress, constipation, failure to thrive, abdominal distention	Complete	CXR	---	---		---

BE: Barium Enema; CT: Computerized tomography; CXR: Chest X-Ray; DE: Diaphragmatic eventration; KUB: Kidney, Ureter, and Bladder X-Ray; MRI: Magnetic resonance imaging.

ultrasound or an abdominal CT scan. Identifying haustra or plicae circularis between the liver and the diaphragm can distinguish pneumoperitoneum from Chilaiditi syndrome.

The majority of the cases with Chilaiditi syndrome require a conservative therapy which includes bed rest in a supine position, daily maintenance bowel regimen with laxatives and normal fiber diet, frequent bowel cleansing, fluid supplementation, and nasogastric decompression[23,25]. In some specific cases emergency surgery may be required: associated volvulus, internal hernia, or acute intestinal obstruction[7,9,22,30,31]. Cases who have lacked the aforementioned surgical conditions and continue to have intractable abdominal pain and respiratory distress may benefit from undergoing a colopexy[6,9,23]. Colopexy is a surgical procedure which involves repositioning of the colon to adhere to the abdominal wall. In our literature review, 21

of the 30 reported cases were managed with a conservative approach and 8 required a surgical intervention (3 had associated volvulus, 4 presented with persistent respiratory distress, and 2 with recurrent vomiting). And of those 8 cases that required surgery, 2 were transverse colectomies, 2 were colopexies, 1 was a colopexy with transverse colectomy, 1 was detorsion, and 2 involved correction of diaphragmatic eventration and elevation of the right hemidiaphragm (Table 1). Of the 30 cases with reported outcomes, the final outcome was satisfactory for all those cases regardless of the treatment approach[6,7,9,22,23].

The teaching point of this uncommon but intriguing syndrome is to have a high index of suspicion of this condition in patients who have predisposing factors. In addition, it is essential to exclude pathologic conditions such as pneumoperitoneum, subphrenic abscess, posterior hepatic lesions, and Morgagni hernia, which can mimic Chilaiditi sign on a radiologic film. A subphrenic abscess usually features a comparatively smaller air fluid level in the right upper quadrant often associated with pleural effusions and basilar atelectasis (this last two conditions not commonly seen with Chilaiditi sign), if the diagnosis is unclear, an abdominal CT scan is recommended for further evaluation[3,23]. In patient with cirrhosis (in the absence of ascites), the prevalence of Chilaiditi sign has been reported to be between 5% and 20%, higher than the general population[31,32]. It is essential to recognize Chilaiditi syndrome particularly in medical procedures requiring percutaneous transhepatic approach such as percutaneous liver biopsy, percutaneous transhepatic cholangiography, or biliary drainage. Real-time ultrasound guide during these procedures can prevent the intestinal injury before the percutaneous access to the liver[33].

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## CONCLUSION

Chilaiditi syndrome is a rare condition especially among the pediatric population. It should be suspected when patients present with constipation, abdominal pain (particularly located in the right upper quadrant), nausea, vomiting, abdominal distention, and respiratory distress of unknown cause. In the cases previously reported, there were no data about recurrence or timeline from first symptomatology to diagnosis; given the lack of information, long-term follow-up in these cases is necessary. In the pediatric population, both conservative and surgical approaches in treating Chilaiditi syndrome, with treatment of the predisposing factors, have resulted in satisfactory outcomes.

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