A Rare Etiology in a Pediatric Patient Presenting with Chronic Cough: Chilaiditi Sign

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ABSTRACT Chilaiditi's sign is a radiological finding characterized by the presence of air below the right hemidiaphragm due to the interposition of a portion of the colon or small intestine between the liver and the diaphragm. It is characterized by the presence of free air beneath the right hemidiaphragm. Chilaiditi sign is a radiological diagnosis. It is largely associated with an anomaly of the falciform ligament in the liver. Patients with this condition typically exhibit gastrointestinal symptoms or respiratory symptoms due to diaphragmatic irritation. This article presents a case study of a child who presented with chronic cough and was diagnosed with Chilaiditi sign.

Keywords: Chilaiditi sign; cough; child; anatomic variation

Chilaiditi sign is a benign variation characterized by the interposition of a segment of the large or small intestine between the liver and the diaphragm.¹ This variation primarily stems from the absence, laxity, or elongation of the falciform ligament, which typically prevents the colonic loops from entering the space between the diaphragm and the liver.²

In children, factors contributing to its etiology include a smaller liver size, degeneration of diaphragmatic muscles, phrenic nerve palsy, pathologies causing increased thoracic pressure, congenital colonic malposition or malrotation, colonic distension due to aerophagia, or constipation.³

First described in 1910 by Demetrius Chilaiditi, a Greek radiologist, this syndrome was identified when he reported three cases of patients exhibiting intra-abdominal free air on radiological imaging due to bowel interposition between the right hemidiaphragm and liver.¹

The incidence ranges from 0.025% to 0.28%, with a higher prevalence observed in males.⁴ Patients typically present symptoms such as abdominal pain, anorexia, vomiting, bloating, constipation, respiratory distress, and chest pain.^{3,4} Radiologically, Chilaiditi syndrome, a diagnosable condition, is recognized by the presence of bowel gas under the right diaphragm on X-rays or Computed Tomography scans.^{1,4}

This article will present a case study of an individual investigated due to chronic cough, revealing the presence of Chilaiditi sign.

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Correspondence: Nazmi Mutlu KARAKAŞ Gazi University Faculty of Medicine, Department of General Pediatric, Ankara, Türkiye E-mail: nmkarakas@gazi.edu.tr Peer review under responsibility of Turkiye Klinikleri Journal of Case Reports. Received: 25 May 2024 Accepted: 26 Dec 2024 Available online: 14 Feb 2025 2147-9291 / Copyright © 2025 by Türkiye Klinikleri. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

CASE REPORT

An 8-year-old girl presented at the pediatric outpatient clinic with complaints of chronic cough and recurrent lower respiratory tract infections. During physical examination, her body temperature was 36.6°C, pulse rate 97 beats per minute, respiratory rate 22 breaths per minute, blood pressure 102/63 mmHg, oxygen saturation 99%, and crepitant rales were heard in the lower right lung fields. Abdominal examination revealed no tenderness, guarding, rebound tenderness, or hepatosplenomegaly. Percussion in the right upper quadrant of the abdomen was tympanic. Laboratory findings showed a hemoglobin level of 12.9 g/dl, white blood cell count of 11,600/uL, absolute neutrophil count of 6,500/uL (56%), and a C-reactive protein level of 10.2 mg/L. Kidney and liver function test results were within normal ranges. Total immunoglobulin levels were within the normal range for her age. Chest X-ray revealed an increase in bilateral bronchovascular markings and radiolucency in the right subphrenic region. This appearance was interpreted as the interposition of a segment of the colon between the liver and the diaphragm. The image was assessed as indicative of Chilaiditi sign (Figure 1a, b). Due to the absence of gastrointestinal symptoms such as abdominal pain, constipation, ileus, or abdominal distension, advanced abdominal imaging was not conducted for the patient.

Written consent was obtained from the patient's parents for the case presentation.

DISCUSSION

Chilaiditi sign is a radiological finding characterized by the presence of air below the right hemidiaphragm due to the interposition of a portion of the colon or small intestine between the liver and the diaphragm. When it causes gastrointestinal symptoms, it is defined as Chilaiditi syndrome.¹⁻³ Patients may present with gastrointestinal complaints such as nausea, vomiting, loss of appetite, abdominal pain, bloating, as well as respiratory symptoms like shortness of breath, cough, and chest pain.³

Chilaiditi syndrome is a benign condition, and in most cases, a conservative approach is sufficient for treatment. In symptomatic patients, fluid and electrolyte support, bowel decompression with a nasogastric tube, and the use of laxatives and enemas are commonly employed.^{5,6} Surgical intervention is not performed in every case. Surgery is indicated in instances unresponsive to conservative treatment, or when associated with rare complications such as cecal perforation, peritonitis, appendicitis, or mesenteric ischemia.⁷ Surgical procedures may include colopexy or peritoneal abrasion of the diaphragm and liver.^{6,7}

Differential diagnoses for Chilaiditi syndrome include pneumoperitoneum, diaphragmatic hernia, subdiaphragmatic abscess, colon perforation, intesti-

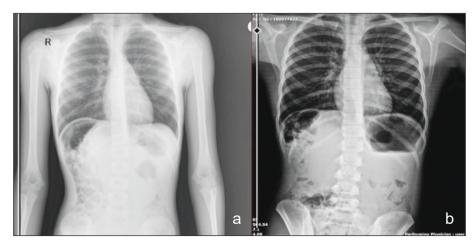


FIGURE 1: Images of the colon loop in the right subdiaphragmatic region.

nal obstruction, intussusception, volvulus, and ischemic bowel. The primary aim is to differentiate between free subdiaphragmatic air and intraluminal air.⁶ A literature review conducted by Gundapenami et al. reported 30 pediatric cases with Chilaiditi sign, with 63% being male and an average age of 4.5 years.⁷ The study also noted colon interposition in 90% of the cases.⁷

In this case, the Chilaiditi sign was an incidental finding. The diagnosis was made after a chest X-ray was taken due to the patient's complaint of cough, which revealed air in the right subdiaphragmatic region. Chronic cough was associated with increased frequency of infection due to diaphragmatic irritation and compression. Additionally, in these patients, the presence of an enlarged colonic loop may cause diaphragm elevation, which is thought to reduce intrathoracic volume and lung ventilation, potentially leading to respiratory symptoms.8 Similar cases have been reported in the literature.^{9,10} In a study by Erdem et al., two pediatric cases with Chilaiditi sign were reported, who were examined due to respiratory distress.9 In the first case, inhaled steroids were administered, while in the second case, treatment for pneumonia with antibiotics and anti-reflux therapy was given. It was reported that acute symptoms subsided with these treatments.9 In a study by Caicedo et al., a case of Chilaiditi syndrome was diagnosed incidentally in a patient under intensive care due to acute respiratory distress.¹⁰ In this case report, the patient did not benefit from conservative treatment and underwent surgery, after which no signs of respiratory failure were observed postoperatively.¹⁰ In our patient, there was a decrease in cough symptoms following supportive treatment, and no additional medical treatment was necessary.

Chilaiditi sign has been associated with neuropsychiatric pathologies like intellectual disability and schizophrenia, leading to a higher incidence of this sign in these populations.¹¹ While its etiology remains incompletely understood, the higher incidence in this patient group might be related to the use of neuropsychiatric medications.¹²

The existing literature advocates for the utilization of abdominal imaging in patients diagnosed with Chilaiditi syndrome who present with gastrointestinal symptoms or signs of intestinal perforation or ischemia. However, in this case, due to the absence of gastrointestinal complaints and lack of clinical signs of intestinal perforation or ischemia, abdominal imaging was not initially. Since gastroesophageal reflux may contribute to cough etiology, further investigations, including esophagogastroduodenal barium swallow and pH monitoring, were planned for differential diagnosis. Our patient is being followed up by the pediatric gastroenterology department for possible gastrointestinal symptoms and differential diagnoses. No organic disease was identified in relation to recurrent lung infections, and follow-up by pediatric pulmonology continues.

Chilaiditi sign is a rare condition that should be considered in the differential diagnosis of patients presenting with unexplained gastrointestinal or respiratory complaints. It should be kept in mind that Chilaiditi sign may also be the cause of unexplained chronic cough.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Hasan Tahsin Çakır, Nazmi Mutlu Karakaş; Design: Hasan Tahsin Çakır, Nazmi Mutlu Karakaş; Control/Supervision: Nazmi Mutlu Karakaş; Data Collection and/or Processing: Selin Kuzucu, Nursima Kunt Baykal; Analysis and/or Interpretation: elin Kuzucu, Nursima Kunt Baykal; Literature Review: Selin Kuzucu, Nursima Kunt Baykal, Hasan Tahsin Çakır; Writing the Article: Selin Kuzucu, Nursima Kunt Baykal, Hasan Tahsin Çakır, Nazmi Mutlu Karakaş; Critical Review: Selin Kuzucu, Nazmi Mutlu Karakaş; References and Fundings: Selin Kuzucu, Nazmi Mutlu Karakaş; Materials: Selin Kuzucu, Nazmi Mutlu Karakaş, Nursima Kunt Baykal, Hasan Tahsin Çakır.

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