

CASE REPORT

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Chilaiditi Syndrome After Colonoscopy: A Case Report and Review of the Literature

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ABSTRACT Chilaiditi Syndrome (CS) was first described by Demetrius Chilaiditi in 1910 as a condition characterized radiologically by the positioning of the colon between the liver and the diaphragm. This condition of unknown cause is very rare in the general population. Although it is usually asymptomatic, it may present with acute, chronic, or recurrent forms and might be associated with symptoms such as constipation, dyspnea, loss of appetite and chest pain. Diagnosis of CS is important since it might be confused with pneumoperitoneum, may cause intestinal obstruction, perforation, and ischemia, and might possibly result in intestinal perforations during colonoscopy. We report the case of a 62-year-old woman who presented with sudden dyspnea and abdominal pain after colonoscopy and was diagnosed as CS after radiological imaging.

Keywords: Chilaiditi Syndrome; colonoscopy; abdominal pain

Chilaiditi Sign (CSign) is a condition where the colon, or more rarely a small bowel segment, is interposed between the liver and the diaphragm. The incidence in the general population is 0.025-0.28%. The incidence increases with age and the male/female ratio is 4/1.¹ It is often asymptomatic and usually diagnosed incidentally. This condition is defined as CSign.² However, it may present with various symptoms such as loss of appetite, abdominal pain, constipation, hiccups, nausea, vomiting, dyspnea, cough, chest pain, and renal colic pain and these symptoms may be acute, chronic, or recurrent. It is defined as CS when it is symptomatic.¹ In light of literature findings, we present the case of a 62-year-old asymptomatic female patient who was evaluated for iron deficiency anaemia. She developed sudden dyspnea, chest pain and abdominal pain following

colonoscopy and was diagnosed with CS following radiological imaging.

CASE REPORT

The 62-year-old female patient was presented to the outpatient clinic with complaints of fatigue, and dizziness. The patient's past medical history was significant for hypertension, allergic asthma, and chronic ischemic heart disease. She had no history of the placement of a cardiac stent or previously performed coronary by-pass. Her family history was unremarkable and there was no history of alcohol or smoking in the patient's social history. On initial evaluation, the patient's vital signs were stable. She was on acetylsalicylic acid, angiotensin converting enzyme inhibitor, inhaler steroid, inhaler selective beta2

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adrenergic receptor agonist and a systemic antihistaminic treatment. On physical examination, she had pallor in the skin and conjunctiva and had occasionally mild bilateral sibilant rhoncus. Laboratory tests were as follows: Haemoglobin: 5.4 g/dl, Hematocrit: 20%, MCV: 67.1 [μ] l, Fe: 8 mg/dl, TIBC: 450 mg/dl, Ferritin: 2.2 mg/dl, B12: 450 pg/ml, Folic acid: 10 ng/mL, renal function tests, liver function tests, amylase, lipase, coagulation values and complete urinalysis were within normal limits. The patient was referred to the hospital with the preliminary diagnosis of iron deficiency anaemia for further evaluation and treatment.

Intravenous erythrocyte suspension and iron carboxymaltose treatment were administered. After the level of haemoglobin increased to 10.6 g/dl, gastroscopy and colonoscopy were performed. There was no complication during gastroscopy but the patient's general condition deteriorated in the operating room after the colonoscopy procedure had been completed. She developed severe dyspnea and abdominal pain. Her oxygen saturation in room air was 80%, blood pressure was 130/90 mmHg, pulse was 95/min, temperature was 36.8 degrees C and fingertip blood sugar was 180 mg/dl. Nasal oxygen therapy was started through a mask, then she was monitored. Electrocardiogram (ECG) showed a ST-segment depression in leads V5-V6. Cardiac enzymes were within normal limits. Arterial blood gas taken under nasal oxygen treatment was normal. The patient had severe abdominal pain. Physical examination revealed no abdominal guarding or rebound tenderness; however, she had extensive tenderness in the abdomen. The preliminary diagnosis was organ perforation and her oral intake was stopped. Direct abdominal radiography in the erect position and posteroanterior chest radiographs were obtained. Colonic loops were observed in the right hemithorax (Figure 1). Oral and intravenous contrast-enhanced thoracic and whole abdominal tomography was obtained. In the absence of intraabdominal free air and oral contrast leakage, the diagnosis of organ perforation was overruled. It was seen in the imaging studies that the stomach was partially in the right hemithorax together with the colonic loops (Figure 2). The right diaphragm was elevated. However, diaphragm integrity could not be evaluated clearly on any

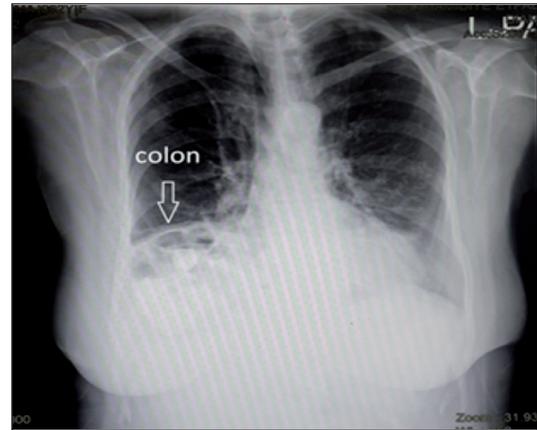


FIGURE 1: Posteroanterior chest X-ray shows elevated right diaphragm and colonic loops in the right hemithorax.

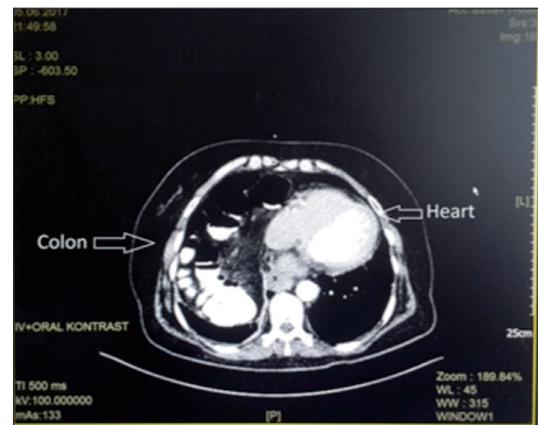


FIGURE 2: Colonic loops appear to be in the right hemithorax.

section. In addition, the patient was diagnosed with CS because the diaphragm integrity was intact and colonic loops were seen just below the diaphragm in the right chest lateral radiograph taken the next day (Figure 3). In the post-procedural follow-up period, the patient was able to pass gas and abdominal pain, dyspnea and ECG findings were regressed. The patient's general condition and vital signs were stable; therefore, oral intake was started one day after colonoscopy. The patient was discharged with full recovery without the need for surgical intervention.

Written informed consent has been obtained from the patient for publishing this case.

DISCUSSION

The colon may be permanently or temporarily interposed between two organs in the abdomen. If these

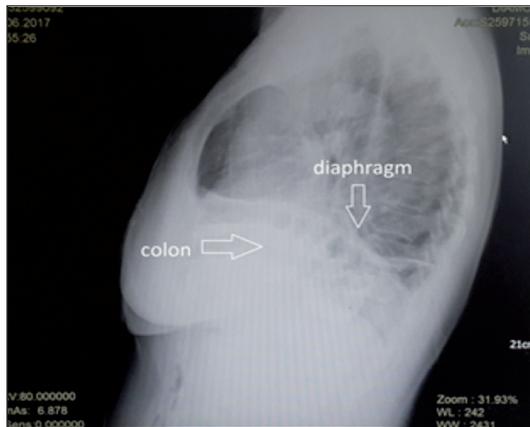


FIGURE 3: Lateral chest X-ray demonstrates an intact diaphragm with colonic loops below the diaphragm.

two organs are the diaphragm and the liver, this is called “hepatodiaphragmatic interposition of the colon” (CSign). The diagnosis of CSign is made by the appearance of air under the right diaphragm with the colonic haustrations. It is an important finding that the localization of the air seen on the radiographs is not changed with the positioning of the patient. Tomography can be used to confirm the diagnosis.³ Demetrius Chilaiiditi, a physician from Istanbul was the first to describe the condition.⁴ Displacement of the colon between any two organs other than the liver and the diaphragm is called a non-Chilaiiditi sign. It is a rare condition. It is usually asymptomatic and is diagnosed incidentally by radiological imaging.⁵

CS was shown at male predominance.¹ It can also be seen in childhood starting from infancy and may present in various clinical forms such as abdominal pain, vomiting, constipation or respiratory distress as in adults.⁶⁻⁸

There are some predisposing factors that increase the risk of CSign. These factors may be due to muscular degenerations or phrenic nerve damage which may cause the diaphragm to be elevated and be above its normal localization, and cirrhosis, right lobe segmental agenesis or relaxation of the suspensory ligament that may lead to a decrease in the liver volume.^{9,10} Colon-related causes (increased colonic mobility, malrotation of the bowel or congenital misalignment) or conditions such as chronic lung disease, pregnancy, ascites, obesity, and aerophagia may cause CSign formation.^{1,11}

CS is usually manifested by gastrointestinal symptoms such as loss of appetite, abdominal pain, constipation, nausea, and vomiting. However, there are case reports in the literature stating that, although rarely, CS may present with recurrent episodes of respiratory distress, typical angina-like chest pain radiating to the left arm, heart failure or a clinical picture mimicking renal colic.^{1,12-14} There are rare cases of CS due to iatrogenic causes in the literature. Apart from our case, only two cases of CS developing as a colonoscopy complication were reported previously.^{1,15}

CS should be differentiated from pneumoperitoneum or diaphragmatic hernias. Free air under the diaphragm may be misdiagnosed as bowel injury and might result in unnecessary surgery.³ Pneumoperitoneum usually occurs after an impact and often requires surgical intervention. In the X-ray, the position of the air can be changed with the position, but there are crescent-shaped gas shadows under the diaphragm and no colonic haustrations are seen.¹⁶ A case report in the literature revealed that a patient who was operated on with the preliminary diagnosis of diaphragmatic hernia after trauma was found to have an intact diaphragm. Thus, computed tomography was obtained and the diagnosis of CSign was made.¹⁷ In addition, there is a case report with a preliminary diagnosis of CS which was subsequently operated with the diagnosis of symptomatic Morgagni hernia.¹⁸ Bowel obstruction, ischemic bowel, volvulus, invagination, appendicitis, and diverticulitis should be considered in the differential diagnosis; however, these conditions may rarely be seen concomitantly with CSign.¹¹

The prevalence of CSign has been higher than 0.3% in plain chest radiographs. This rate is 2.4% in thoracic and abdominal tomography.¹⁹ There is no need for surgical intervention in asymptomatic cases with CSign. Treatment usually involves bed rest, laxatives, enemas, nutritional support with a diet high in fibre and intravenous fluid therapy. Although a conservative approach is generally enough, treatment is surgery in emergency cases such as intestinal obstruction, volvulus, intestinal ischemia, or perforation.^{1,3,16} In patients with chronic complaints, elective surgery may be considered to prevent possible complications.²⁰ In our case, the patient’s complaints re-

gressed after supportive treatment alone following colonoscopy.

We have seen an increase in the number of CSign cases in the literature in recent years due to the increase in the frequency of use of imaging methods for diagnosis of various conditions. This case is one of the rare cases in the literature due to its development as an acute colonoscopy complication. It should be kept in mind that the presence of a CSign which is previously unknown might cause life-threatening consequences after an invasive procedure such as colonoscopy in patients with predisposing factors.

Source of Finance

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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: Fatih Borlu; **Design:** Dilek Toprak; **Control/Supervision:** Güzin Zeren Öztürk, Dilek Toprak; **Data Collection and/or Processing:** Yağmur Gökseven; **Analysis and/or Interpretation:** Yağmur Gökseven, Güzin Zeren Öztürk, Fatih Borlu; **Literature Review:** Yağmur Gökseven; **Writing the Article:** Yağmur Gökseven; **Critical Review:** Dilek Toprak, Güzin Zeren Öztürk, Fatih Borlu.

REFERENCES

1. Yin AX, Park GH, Garnett GM, Balfour JF. Chilaiditi syndrome precipitated by colonoscopy: a case report and review of the literature. *Hawaii J Med Public Health.* 2012;71(6):158-62. [PubMed]
2. Hountis P, Chounti M. Chilaiditi's sign or syndrome? Diagnostic question in two patients with concurrent cardiovascular diseases. *Monaldi Arch Chest Dis.* 2017;87(2):775. [Crossref] [PubMed]
3. Mistry KA, Bhoil R, Kumar N, Chopra R, Thapar N. Significance of Chilaiditi sign and Chilaiditi syndrome. *Emerg Nurse.* 2017;5(3):26-31. [Crossref] [PubMed]
4. Chilaiditi D. Zurfrage der hepatoptose und pto-seimallgemeinenimanschluss an dreifälle von temporärer, partieller Leberverlagerung. *Fortschr Geb Rontgenstr.* 1910;16:173-8.
5. Bredolo F, Esposito A, Casiraghi E, Cornalba G, Biondetti P. Intestinal interposition: the prevalence and clinical relevance of non-hepatodiaphragmatic conditions (non-Chilaiditi forms) documented by CT and review of the literature. *Radiol Med.* 2011;116(4):607-19. [Crossref] [PubMed]
6. Çetinkaya E, Razi CH, Gündüz M. Chilaiditi's syndrome: a case report. *Türkiye Klinikleri J Pediatr.* 2004;13(1):33-6.
7. Correa Jiménez O, Buendía De Ávila M, Parra Montes E, Davidson Córdoba J, De Vivero Camacho R. Chilaiditi's sign and syndrome: rare conditions but diagnostically important in pediatrics. *Clinical cases. Rev Chil Pediatr.* 2017;88(6):828. [Crossref] [PubMed]
8. Huang WC, Teng CS, Tseng MH, Lin WJ, Wang CC. Chilaiditi's syndrome in children. *Acta Paediatr Taiwan.* 2007;48(2):77-83. [PubMed]
9. Ogasawara M, Ishiyama A, Sugiura A, Segawa K, Nonaka I, Takeshita E, et al. Duchenne muscular dystrophy with platypnea-orthodeoxia from Chilaiditi syndrome. *Brain Dev.* 2018;40(4):339-42. [Crossref] [PubMed]
10. Krishna P, Panda PK, Hariprasad S, Singh SS, Gedela SR. A case of liver cirrhosis and Chilaiditi syndrome with atypical pneumonitis. *J Family Med Prim Care.* 2018;7(2):471-4. [Crossref] [PubMed] [PMC]
11. Moaven O, Hodin RA. Chilaiditi syndrome: a rare entity with important differential diagnoses. *Gastroenterol Hepatol (N Y).* 2012;8(4):276-8. [PubMed]
12. Bulut Y, Yılmaz N, Memur Ş, Altınöz S, Öztürk A. [A case with recurrent respiratory distress: Chilaiditi syndrome]. *Çocuk Dergisi.* 2008;8(4):243-6.
13. Sorrentino D, Bazzocchi M, Badano L, Toso F, Giagu P. Heart touching Chilaiditi's syndrome. *World J Gastroenterol.* 2005;11(29):4607-9. [Crossref] [PubMed] [PMC]
14. Okiro JO. Chilaiditi syndrome mimicking congestive heart failure. *BMJ Case Rep.* 2017 Jul 14. Doi: 10.1136/bcr-2017-220811. [Crossref] [PubMed] [PMC]
15. Bernui G, Tagle M. Chilaiditi's syndrome after colonoscopy. *Clin Gastroenterol Hepatol.* 2020;18(1):e3. [Crossref] [PubMed]
16. Weng WH, Liu D, Feng CC, Que R. Colonic interposition between the liver and left diaphragm-management of Chilaiditi syndrome: a case report and literature review. *Oncol Lett.* 2014;7(5):1657-60. [Crossref] [PubMed] [PMC]
17. Kamiyoshihara M, Ibe T, Takeyoshi I. Chilaiditi's sign mimicking a traumatic diaphragmatic hernia. *Ann Thorac Surg.* 2009;87(3):959-61. [Crossref] [PubMed]
18. Vallee PA. Symptomatic morgagni hernia misdiagnosed as Chilaiditi syndrome. *West J Emerg Med.* 2011;12(1):121-3. [PubMed]
19. Prassopoulos PK, Raissaki MT, Gourtsoyianis NC. Hepatodiaphragmatic interposition of the colon in the upright and supine position. *J Comput Assist Tomogr.* 1996;20(1):151-3. [Crossref] [PubMed]
20. Koşar A, Tezel Ç, Örki A, Kırıl H, Ürek Ş, Dudu C. [Hepatodiaphragmatic interposition: Chilaiditi's syndrome: case report]. *İzmir Göğüs Hastanesi Dergisi.* 2004;18(3):133-5.