CASE REPORT

Chilaiditi's syndrome complicated by colon perforation: a case report

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ABSTRACT

Hepatodiaphragmatic interposition of the small or large intestine is known as Chilaiditi syndrome, whichis a rare disease diagnosed incidentally. Chilaiditi syndrome is typically asymptomatic, but it can be associated with symptoms ranging from intermittent, mild abdominal pain to acute intestinal obstruction, constipation, chest pain and breathlessness.A 54-year-old male patient was admitted to the hospital with a history of abdominal pain, nausea and vomiting. Chest X-ray revealed an elevation of the right hemidiaphragma caused by the presence of a dilated colonic loop below.The patient underwent urgent surgery with perforation as preliminary diagnosis.The patient underwent right hemicolectomy and ileocolic anastomosis because of the intestinal obstruction related to Chilaiditi's Syndrome. Due to the rarity of this syndrome and typical radiological findings, this case was aimed to be presented.

Key words: Abdominal pain; Chilaiditi's syndrome; surgery.

INTRODUCTION

Interposition of the bowel (usually transverse colon or hepatic flexura) or the small intestine between the liver and diaphragm, which is a rare anomaly, was first defined by the Greek radiologist Demetrius Chilaiditi in 1910.[1,2] It is incidentally seen 0.025–0.28% in the general population.[3] Its incidence increases with advancing age and it is seen rarer in children when compared to adults. It is more frequently seen in male patients. Chilaiditi syndrome can cause a variety of symptoms including abdominal pain, nausea, vomiting, and small bowel obstruction. Specific symptoms and presentation of Chilaiditi syndrome can vary greatly from one person to another. The cause of Chilaiditi syndrome is not fully understood.

The objective of this study was to report a case that presented with intestinal obstruction caused by Chilaiditi syndrome and review the relevant literature.

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CASE REPORT

A 54-year-oldmale patient was admitted to the Emergency Department of Surgery, Izmir Katip Celebi University Ataturk Training and Research Hospital with a 24-hour history of right upper abdominal pain, nausea and vomiting. Although the severity had been altering, these complaints had persisted for 6 months. Physical examination revealed epigastric and right upper abdominal tenderness and rigidity; no rebound tenderness was identified. In addition, auscultation revealed hypoactive bowel sounds. Complete blood count and blood biochemistry were normal except hipoalbuminemia (2,1 g/dL) and leukocytosis (17000 mm/dl). Plain chest radiography demonstrated the elevation of the right diaphragm and right subdiaphragmatic air (Fig. 1). Abdominal computed tomography (CT) scan reported dilatation related to gastric outlet obstruction, dilatated small bowel loops and hepatic flexura, intraperitoneal free air and fluid, which may be related to perforation (Figs. 2a, b). According to these findings, the patient underwent urgent surgery with a preliminary diagnosis of perforation. During exploration, the stomach was lying to pelvis, and advanced dilatation of the small intestines and stomach was observed. Besides, ascending and transverse colon was located between the liver and diaphragm, and segmental necrosis and focal microperforations were seen. The patient underwent right hemicolectomy and ileocolic anastomosis because of the intestinal obstruction related to Chilaiditi syndrome (Fig. 3). Starting from the ninth day, the patient defecated. His postoperative course was uneventful and was discharged on the tenth day. The patient was followed up for four months and remained asymtomatic.

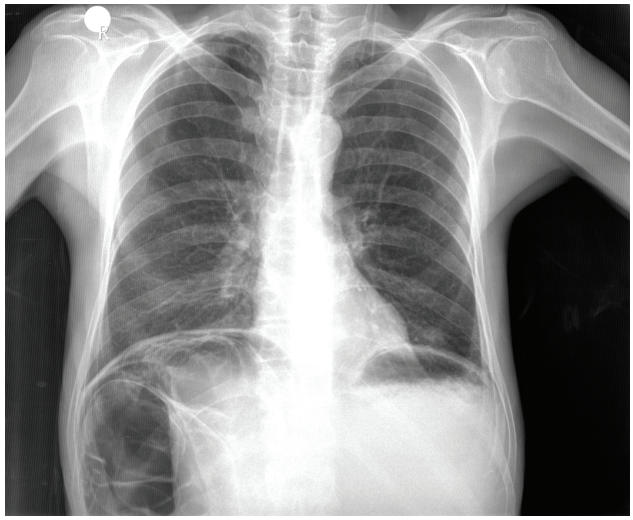

Figure 1. The elevation of the right hemidiaphragm and air under the diaphragm draw attention to the anterior-posterior chest X-ray.

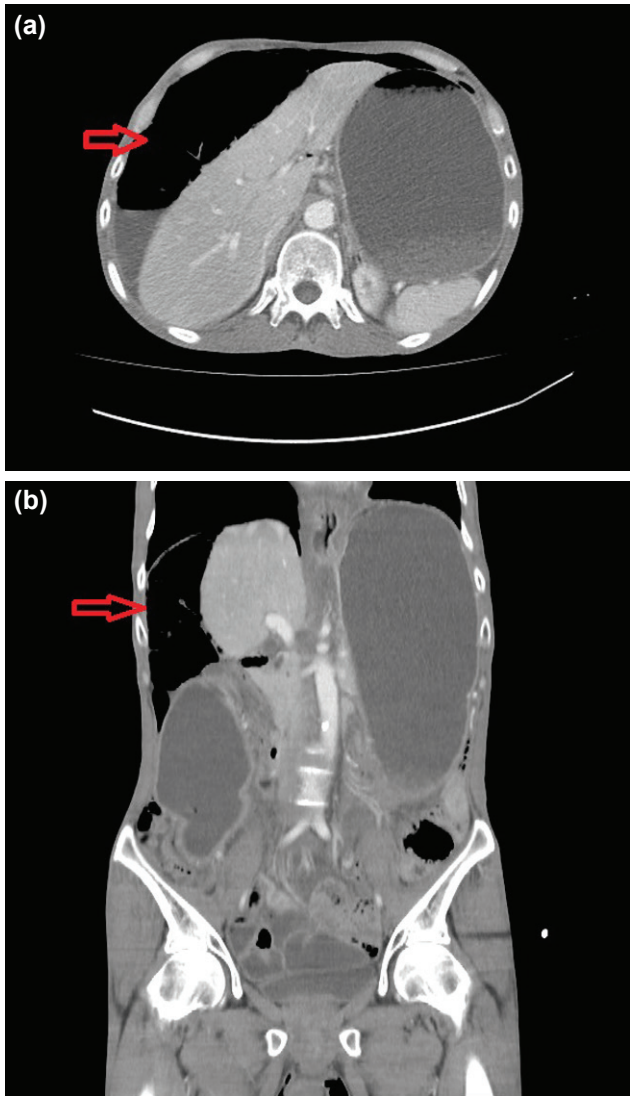

Figure 2. (a) Axial CT / **(b)** Coronal CT: Intestinal loop transposed between the diaphragm and liver (indicated by the arrow).

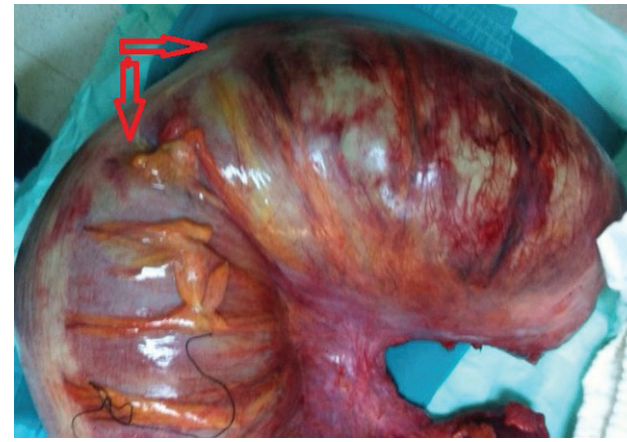

Figure 3. Intra-operative pictures of a bowel microperforation (indicated by the arrow).

DISCUSSION

Intestinal interposition is a medical condition where a segment of the bowel is temporarily or permanently interposed between two organs. Among these, the hepatodiaphragmatic interposition is termed Chilaiditi sign. Chilaiditi sign is usually asympomatic, and when accompanied by clinical symptoms, it is termed as Chilaiditi syndrome, whichis an extremely rare disorder. Normally, the suspensory ligament of the liver, mesocolon, liver, and the falciform ligament limit the surrounding space around the liver and prevent colonic interposition. Chilaiditi syndrome may be congenital or acquired. It can be caused by obesity, multiple pregnancies; liver related problems such as ptotic or small liver, cirrhosis; diaphragmatic problems such as degeneration of diaphragm muscles, phrenic nerve paralysis, tuberculosis or increased intrathoracic pressure caused by emphysema; colonic factors such as anormal enlargement of colon, suspensory ligament abnormality or agenesis and congenital malposition or malrotation.[4–6]

Median age of the patients at presentation was 60 years in the literature. Male to female ratio was 4:3.[2,3] Chilaiditi syndrome is most commonly seen in the elderly with a cadence of 1%, but there have been cases where it was presented in patients as young as 5 months in the literature.[6,7] Our patient was a 54-year-old male who corresponded to the information in the literature.

Most patients do not have any complaints, and the disorder is detected in radiological examinations incidentally.However, it can cause acute, chronic or repetitive digestive complaints such as abdominal pain, vomiting, constipation, swelling, anorexia, respiratory distress, and chest pain. Moreover, it may cause situations such as volvulus, incarceration, and perforation that require urgent surgical intervention.[8,9] There is a high risk of perforation during liver biopsy or colonoscopy in patients who have not been diagnosed.[6] Our patient had intestinal obstruction findings such as severe abdominal pain, vomiting, and distention.

Diagnosis of Chiliaditi syndrome with only history, physical exam and blood work is almost impossible, and it is usually diagnosed after radiological imaging studies. It is diagnosed with routine chest radiography and direct abdomen radiography incidentally. CT and ultrasonography (US) are required for differential diagnosis.[10,11] The first step is to rule out the possibility of pneumoperitoneum. CT plays an important role when performed to differentiate it from perforation. On CT, recognizing the colonic haustra behind the liver is diagnostic for Chilidiati syndrome. US is also useful in differentiating Chilidiati syndrome from pneumoperitoneum, which usually requires an immediate surgical intervention.[2,11] Pleural effusion and atelectasis related to Chiliaditi syndrome may occur. In differential diagnosis, subdiaphramatic rupture, posterior liver lesions and retroperitoneal masses must be considered.

Treatment for Chilidiati syndrome is generally conservative, including bed rest, high fiber diet, intravenous hydration, nasogastric decompression, enema, cathartics, and laxatives.[11,12] However, surgery must be performed to prevent possible complications in patients with chronic complaints, or when complications such as ischemia, perforation, intestinal obstruction are suspected. When the literature is reviewed, complications of Chiliaditi syndrome requiring urgent surgery can be listed as cecal and colonic volvulus, subphrenic appendicitis, internal hernia, and cecum perforation.[1,12–16] Saber and Boros have previously reported that 26% of the patients require operative management.[11] There is no clear consensus on the best surgical approach. A variety of procedures described in the literature include colon resection, hepatopexy, colopexy, right hemicolectomy, sigmoidectomy, and subtotal colectomy.[1,6,8] Otherwise, emergency surgery is performed as was in our case.

In conclusion, Chilaiditi syndrome is usually asymptomatic. It is a rarely considered differential diagnosis with vague symptoms that make the diagnosis difficult. Colonic distention may cause stomachache, vomiting, and shortness of breath. It is mostly diagnosed with X-rayrequested for another reason. Ultrasonography and CT may be required for differential diagnosis. Treatment is usually conservative, but it should be noted that an urgent surgery may have to be performed due to complications. Yet, a proper work-up and keeping in mind the possibility of Chilaiditi syndrome may prevent the patient from undergoing unnecessary surgery.

Conflict of interest: None declared.

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OLGU SUNUMU - *ÖZET*

Kolon perforasyonu yapan Chilaiditi Sendromu: Bir olgu sunumu

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Chilaiditi sendromu, ince kalın bağırsakların hepatodiafragmatik interpozisyonu durumudur. Nadir görülür ve olguların çoğuna tesadüfen tanı konur. Sıklıkla semptomsuz olmakla birlikte aralıklarla ortaya çıkan hafif abdominal ağrı, intestinal obstrüksiyon, kabızlık, göğüs ağrısı, nefes darlığı gibi semptomlarla da ortaya çıkabilir. Karın ağrısı, bulantı ve kusma öyküsüyle hastaneye başvuran 54 yaşında erkek hastanın çekilen akciğer grafisinde dilate kalın bağırsak ansının alttan basısı sonucu sağ diyafragmanın yükselmiş olduğu gözlendi. Hasta perforasyon ön tanısıyla acil operasyona alındı. Chilaiditi's sendromunun neden olduğu intestinal obstrüksiyon nedeniyle sağ hemikolektomi ve ileokolik anastomoz uygulandı. Nadir görülen bir sendrom olması ve tipik radyolojik bulgular nedeniyle bu olguyu sunmayı amaçladık.

Anahtar sözcükler: Cerrahi; Chilaiditi sendromu; karın ağrısı.

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