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CASE REPORT
ΕΝΔΙΑΦΕΡΟΥΣΑ ΠΕΡΙΠΤΩΣΗ

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Chilaiditi syndrome
A possible diagnostic challenge

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Σύνδρομο Chilaiditi:
μια πιθανή διαγνωστική πρόκληση

Περίληψη στο τέλος του άρθρου

Chilaiditi's sign is characterized by the asymptomatic incidental radiographic finding of an intestinal loop interposed between the liver and the diaphragm; the Chilaiditi syndrome refers to the sign and the associated abdominal or lower chest symptoms. An elderly male with comorbidities was admitted to hospital with a suspected perforating acute abdomen, because computed tomography (CT) showed redundant sigmoid, discrete pneumoperitoneum, distension of intestinal loops and hydroaeric levels. He was lethargic and with a gastrostomy. In the previous 24 hours he had presented nausea, vomiting, diarrhea, diffuse abdominal pain and reduced urine output. The chest X-ray showed air under the diaphragm and marked dilation of colonic loops causing diaphragmatic elevation, whereas abdominal CT confirmed the classic features of the Chilaiditi syndrome. Conservative management, which is the first option, resulted in improvement for this potentially severe entity. Clinicians should be aware of both the classical Chilaiditi sign and syndrome to avoid undue interventions.

Key words

Abdominal pain
Chilaiditi syndrome
Diagnosis
Radiology

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Chilaiditi's sign is characterized by the asymptomatic incidental radiographic finding of an intestinal loop interposed between the liver and the diaphragm, while the Chilaiditi syndrome (CS) refers to those cases associated with abdominal or lower chest symptoms.[1–15] CS is a rare condition, first described in 1910, mainly characterized by abdominal or retrosternal pain, anorexia, vomiting, bowel distension, constipation, and subocclusion.[2,10] This transient or permanent interposition of segments of colon, small intestine (rare), or stomach (very rare) can be identified on chest or abdominal imaging.[4] Considered as a benign entity, when associated with other acute diseases, the incidence of CS on X-rays is estimated at between 0.025% and 0.28% in the general population, but may increase with age, mainly over 60 years, mental disorder, and male gender.[3,4,8] The reported incidence is 0.3% on chest X-ray and 2.4% on chest/abdominal computed tomography (CT).[7]

CS can be detected even in the absence of pain, when images of the chest or abdomen are obtained to investigate other conditions. Predisposing factors include a history of abdominal surgery, constipation, aerophagia, mental disorder, chronic pulmonary disease, obesity, pregnancy, liver cirrhosis and hypothyroidism.[14] The early, prompt diagnosis of CS in patients with abdominal pain is mandatory, as the presence of an intestinal loop between the diaphragm and the liver can propitiate a severe reduction in blood flow. Although a rare condition, with scarce symptoms and without surgical indications, CS should be a hypothesis during the diagnostic evaluation of diverse gastrointestinal disorders: The earliest possible correct identification of CS will avoid unnecessary invasive interventions due to misdiagnosis, and allow the appropriate conservative management.[3,11,12]

The objective of the present case report is to enhance the suspicion about a rarely reported condition that may be occasionally mistaken by a perforated surgical acute abdomen.

CASE PRESENTATION

An 84-year-old male patient with progressive supranuclear palsy, Parkinson's disease, and a previous hemorrhagic stroke was transferred to the emergency unit of the hospital with suspicion of perforated acute abdomen. He was lethargic, disoriented in time and space, answering questions with difficulty, slowly and babbling, and he had a gastrostomy. Abdominal CT showed a redundant sigmoid, pneumoperitoneum at the right pelvis, and distended intestinal loops with hydroaeric levels. In the next 24 hours, he presented nausea, vomiting, intense diffuse abdominal pain, and also tenderness on superficial palpation, liquid stools with hematochezia, dysuria, dark colored urine with foul odor, and a reduced daily urine output. On examination, he was acyanotic,

anicteric, afebrile, sudoreic, hypoactive, pale (+/4+), hypotensive (94/52 mmHg), with tachycardia (104 to 110 bpm); he had a rigid distended abdomen, and accentuated hydro-air sounds, but without pain on palpation. He was using a 12 L/minute non-rebreathing mask, oxygen saturation 99%, and respiratory auscultation showed physiological breath sounds and a respiratory rate of 24 ipm. The radial pulses were rhythmic, symmetrical, and cardiac auscultation showed normal rhythmic, sounds, in two stages and without murmurs. His extremities were well-perfused, with the capillary filling time less than 3 seconds. Laboratory investigation: White blood count: 3,205/mm^3 (rods 0% and segmented 40%); platelets: 180,000/mm^3; magnesium: 1.9 mg/dL; sodium: 138 mmoL/L; urea: 23 mg/dL; potassium: 3.8 mmoL/L; creatinine: 0.6 mg/dL; alanine transaminase (ALT): 15.6 U/L; aspartate transaminase (AST): 20.7 U/L; procalcitonin: 0.059 mg/mL; amylase: 78 U/L; lipase: 35.4 U/L; total bilirubin: 0.27 mg/dL (direct: 0.11; indirect: 0.16); γ-GT: 21 U/L; alkaline phosphatase: 53.2 U/L; creatine phosphokinase (CPK): 86 U/L; C-reactive protein (CRP): 0.57 mg/dL. Urine culture revealed: *Enterobacter cloacae* >100,000 CFU/mL. Chest X-ray showed the presence of air under the diaphragm and marked dilation of colonic loops causing diaphragmatic elevation (fig. 1). Abdominal CT confirmed the classic picture of CS (figures 2A, 2B). The brain CT imaging did not show any acute changes that could justify the clinical picture. A general surgery consultation discarded the need for exploratory laparotomy for diagnostic or treatment purposes.

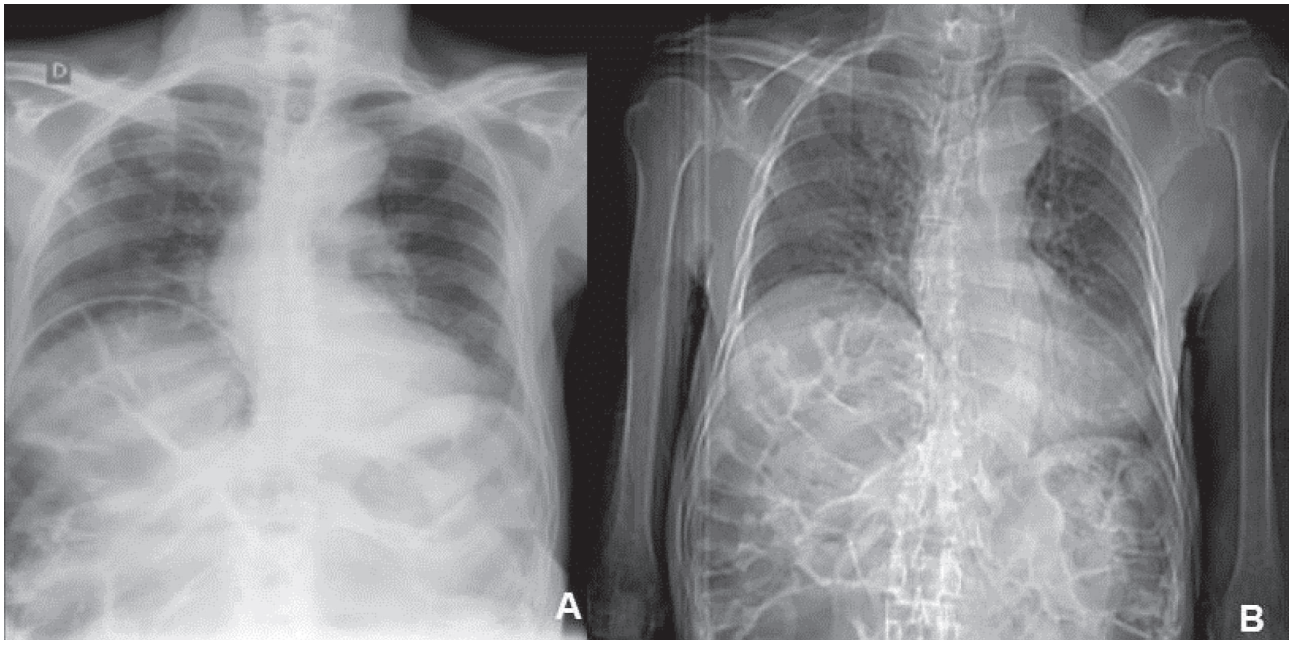

Figure 1. Chilaiditi syndrome in an 84-year-old male: Chest X-ray showing marked dilatation of colonic loops, causing elevation of the right diaphragm (**A** and **B**).

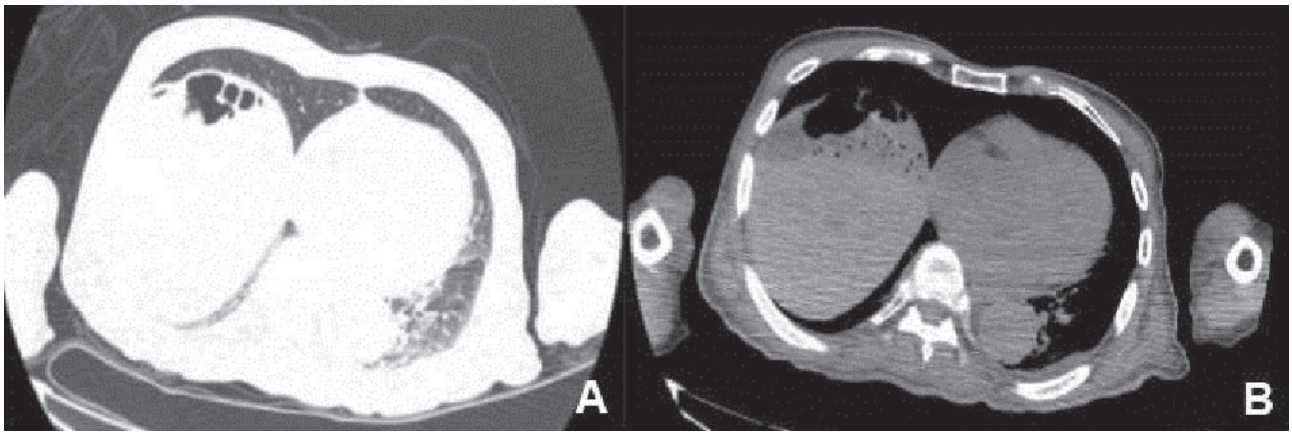

Figure 2. Chilaiditi syndrome in an 84-year-old male: Abdominal computed tomography (CT) confirming the interposition of the colon between the anterior face of the liver and the diaphragm (**A** and **B**).

Due to the patient's status and basic clinical condition, palliative measures were chosen. During the initial care, the patient's clinical condition was stabilized by using 2,000 mL of 0.9% saline solution, with improvement in hypotension. Antiemetic and analgesic medications and empirical antibiotic therapy with intravenous ertapenem and metronidazole were administered by the enteral route. He remained fasting for 12 hours, receiving venous hydration, general condition monitoring, and undergoing antibiotic and analgesic medications with schedules determined for 24 hours. After the suspension of painkillers, there was no worsening of symptoms, and the necessity for emergency surgery was ruled out. He followed the antibiotic therapy for 7 days and showed significant rapid clinical improvement.

DISCUSSION

The elderly male described here had a colonic interposition in the hepatodiaphragmatic space, with manifestations of pseudoperitoneum suggesting acute perforated abdomen, typical of the CS. Misdiagnosis may occur due to the herniation through the right diaphragm of abdominal viscera including the colon, small intestine, and stomach, as along with pneumoperitoneum. In spite of his chronic comorbidities and the challenging differential diagnosis of a severe acute abdominal surgical condition, he was not submitted to invasive procedures that could increase the morbimortality,[15] and the conservative clinical management was successful.[15] This uncommon condition is estimated to account for 0.025% to 0.28% of chest X-ray findings worldwide, with a 4:1 male-to-female ratio, and an incidence of 1.18% and 2.40% on CT.[5,9] Predisposing factors for CS include the absence, laxity or elongation of the suspensory ligaments of the transverse colon, redundant colon, elevation of the right hemidiaphragm, and atrophy or hypoplasia of the liver, which is considered a rare cause of these alterations.[9] Other mechanisms involve great weight loss in the obese people, chronic constipation due to stretching and redundancy of the colon, abnormally elevated diaphragm or diaphragmatic paralysis, congenital malformations, chronic obstructive pulmonary disease (COPD), gas distension of the colon; massive ascites with increased intra-abdominal pressure, and multiple pregnancies. Although usually oligosymptomatic, CS occasionally manifests with an accentuated abdominal pain, nausea, vomiting, constipation, abdominal distention, and sub-occlusion or intestinal obstruction, in addition to respiratory distress, dyspnea, and retrosternal pain.[1,8] Santos and colleagues reported an 85-year-old woman with Ogilvie's syndrome (OS), a condition first described in 1948, that shares clinical and imaging features with CS.[10] OS is classically characterized by an acute intestinal pseudo-obstruction and colonic dilation. Although exceeding rare, elderly people can have associated OS and CS, pneumoperitoneum or pseudopneumoperitoneum.[10] This initial concern of acute abdomen was discarded, based on the imaging findings. The treatment of asymptomatic patients is generally non-invasive, and surgical procedures are indicated only in cases of urgency, or the failure in a conservative management.[6,13] Early recognition of severe complications, such as obstruction, ischemia, or perforation, is essential, for which surgery is indicated, and for cases not responsive to the non-invasive measures.[14] Correct differential diagnosis will avoid unnecessary endoscopic or laparoscopic interventions, lowering the risk of intestinal ischemia and perforation, respiratory failure, or even death.[4,12]

In conclusion, CS can be mistaken for other entities, and publication of reports will allow reduction of undue interventions. These comments aim to increase the knowledge of clinicians about this rarely reported syndrome, focusing on the role of an accurate imaging evaluation of the air under the diaphragm, because if the diagnosis is correct, more effective, conservative treatment can be successful.

ΠΕΡΙΛΗΨΗ

Σύνδρομο Chilaiditi: μια πιθανή διαγνωστική πρόκληση

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Το σημείο Chilaiditi χαρακτηρίζεται από το ασυμπτωματικό ή όχι ακτινογραφικό εύρημα εντερικής έλικας που παρεμβάλλεται μεταξύ ήπατος και διαφράγματος. Το σύνδρομο Chilaiditi αναφέρεται σε συμπτώματα τόσο από την κοιλιά όσο και τη βάση του θώρακα. Ηλικιωμένος ασθενής μεταφέρθηκε στο νοσοκομείο με υποψία διάτρησης και οξεία κοιλία, επειδή σε μια αξονική τομογραφία (CT) εμφανίζονταν διάταση σιγμοειδούς, πνευμοπεριτόναιο, διάτα-

ση εντερικών ελίκων και υδραερικά επίπεδα. Ήταν ληθαργικός και έφερε γαστροστομία. Τις τελευταίες 24 ώρες εμφάνιζε ναυτία, έμετο, διάρροια, διάχυτο κοιλιακό άλγος και μειωμένη παραγωγή ούρων. Η ακτινογραφία του θώρακα έδειξε αέρα κάτω από το διάφραγμα και διάταση εντερικών ελίκων, ενώ η κοιλιακή CT επιβεβαίωσε τα κλασικά χαρακτηριστικά του συνδρόμου. Η συντηρητική αγωγή είχε ως αποτέλεσμα τη βελτίωση αυτής της δυνητικά σοβαρής περίπτωσης. Οι θεράποντες ιατροί πρέπει να γνωρίζουν τόσο το κλασικό σημείο όσο και το σύνδρομο Chilaiditi για την αποφυγή αδικαιολόγητων παρεμβάσεων.

Λέξεις ευρετηρίου: Ακτινολογία, Διάγνωση, Κοιλιακό άλγος, Σύνδρομο Chilaiditi

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