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Two case series of Chiladiti syndrome and a review of literature

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Abstract

Introduction: Chilaiditi's syndrome was repeatedly an unforgettable case in Emergency Department due to its various symptoms.

Methods: This case report consisted two reported cases of Chilaiditi syndrome. Then, we followed the methods of PRISMA for systematic review.

Results: In this report, two cases of Chilaiditi syndrome with the different chief of complaint when admitted to the hospital. A 35-year-old woman presented to the Emergency ward with abdominal pain, while the older subject, a 67-year-old man complained of shortness of breath. Plain radiograph showed an elevated right hemidiaphragm and the presence of subphrenic air, making the diagnosis of Chilaiditi syndrome. Both patients were managed conservatively with nasogastric tube decompression and intravenous fluid. The follow up showed favorable results and symptoms resolved.

Discussion: Chilaiditi syndrome might not be a rare entity, but a difficult syndrome to identify clinically due to its various unspecific symptoms. The radiological examination was needed to be carried out for the diagnosis. Conservative management led to favorable results.

Conclusion: Clinicians should be more aware of the diagnosis of Chilaiditi syndrome with its various symptoms. Radiological evidence should be obtained for the confirmed diagnosis, and conservative management was the modality choice.

Keywords: Chilaiditi syndrome; radiology; conservative.

Introduction

Chilaiditi was first described by a radiologist, named Demetrius Chilaiditi in 1910, after his publication of 3 cases of hepatodiaphragmatic interposition of colon [1]. Chilaiditi's syndrome refers to either temporary or permanent interposition of the small intestine or colon to the hepatodiaphragmatic space, causing symptoms. Male dominated the cases with a 4:1 ratio than that of female [2]

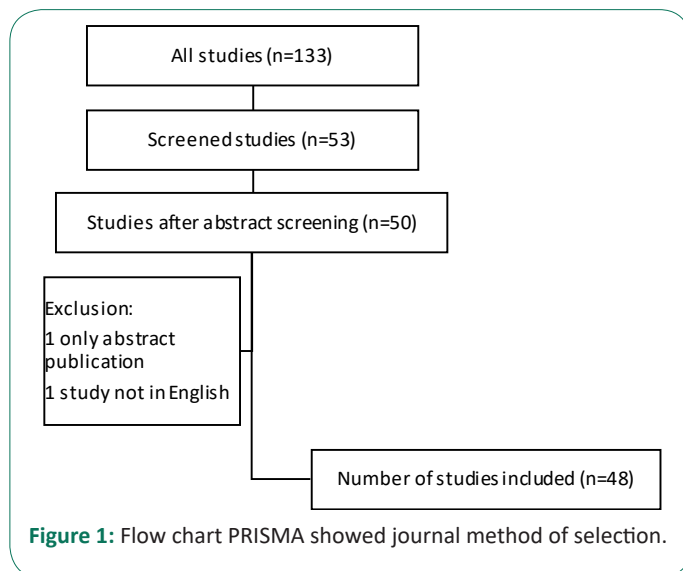
Chilaiditi syndrome was mostly asymptomatic or present with mild signs such as unspecified abdominal pain, nausea, vomiting,

and constipation. In advanced cases, it might be presented with shortness of breath if the lung was lifted [3]. These symptoms could be misled the surgeon to carry out unnecessary surgery for seriously mistaken abnormalities [4]. However, on the other side, this syndrome could precipitate or found coincidence with other serious abnormalities. Regardless of that, in the Emergency Department, the diagnosis of the chilaiditi syndrome was mostly forgotten to be a differential diagnosis of such various symptoms. In this case report, we presented two cases of the chilaiditi syndrome, who presented with different symptoms.

Methods

This case report is described based on two cases encountered in center in the past 1 year. Literature searching for systematic reviews followed the methods of PRISMA. Systematic reviews cover all Chiladiti cases reported throughout the world. The parameters assessed were symptoms, clinical findings, radiological findings, management, and results of treatment. In addition, the results of the systematic review were also compared to the cases we reported in the discussion section.

In a systematic literature search of the Publisher and Cochrane Central Register of Controlled Trials databases using the search terms ("chiladiti" [MeSH Terms] OR "chilaiditi" [All Fields]) AND ("Syndrome" [MeSH Terms] OR "syndrome" [All Fields]), we identified 133 citations defined for the purpose of this review. From the 133 articles, 47 articles consisting of 60 cases were included which were matched with the parameters from this study. The data was managed by PRISMA guidelines and other Note programs. Figure 1 shows a flow diagram of the literature search algorithm.



Results

Case 1

A 35-year-old woman was presented with intermittent epigastric pain for 3 days before admitted to the Emergency Department. The epigastric pain was worse, with the presence of nausea and vomiting since 2 days ago. She also complained about the difficulty of passing stool. She stated that she did not have flatus for 1 day. Any history of surgery and previous illness were denied. Physical examination showed epigastric tenderness without any sign of peritoneal irritation. The patient was firstly evaluated by the primary health care without any imaging modalities, then she was referred to our hospital with the suspicious diagnosis of acute abdomen. Laboratory findings were within normal limits. Then, she underwent abdominal X-Ray, in the supine, erect, and left lateral decubitus position. The plain radiographs findings revealed the presence of air at the subphrenic which did not change in the left lateral decubitus

X-Ray (Figure 2). The right hemidiaphragm was also shown to be elevated. Abdominal ultrasound confirmed the prominent haustra at the right hemidiaphragm, a Chiladiti sign. The patient was managed conservatively with intravenous fluid and conservatively. She was discharged after 2 days of inpatient care with the favorable condition. However, the patient refused any radiograph to be done for final confirmation due to relieving symptoms.

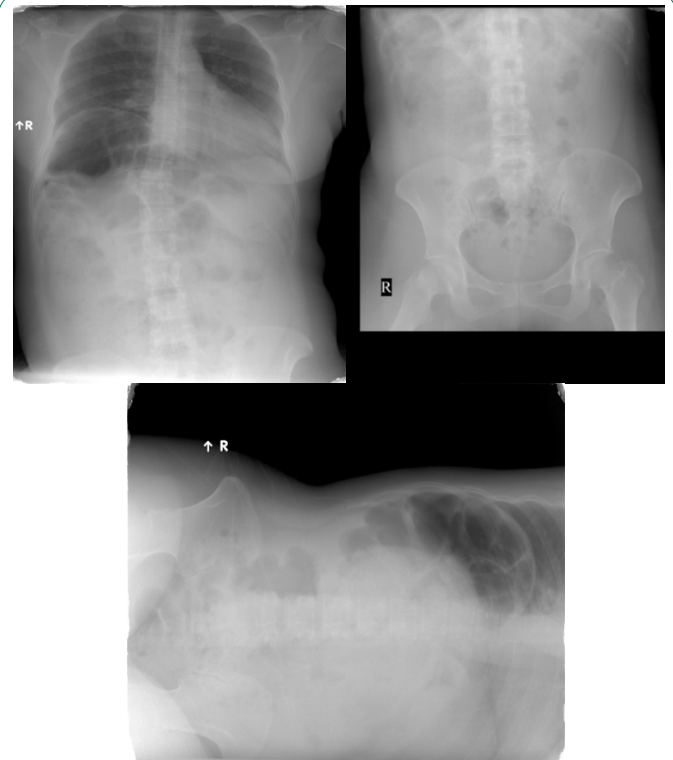


Figure 2: Plan abdominal radiograph in supine, erect, and left lateral decubitus position.

Case 2

A 62-year-old man presented with shortness of breath for 1 week before hospital admission. The patient has a history of mass at the buccal which has not been confirmed by open biopsy. He also complained of unspecified abdominal pain, but did not come too often. He had nausea but denied any history of vomiting. He had additional symptoms, epistaxis, which was suspect from the tumoral bleeding. Physical examination showed only slight decreased breath sound at the bottom of the right lung. Unremarkable findings were shown for the abdominal region. Chest radiograph was obtained and confirmed the elevated right hemidiaphragm, a Chiladiti sign, with the presence of minimal right lung pleural effusion due to pulmonary metastases (Figure 2). He was then managed conservatively with oxygenation, decompression and intravenous fluid. In this patient, he also received tranexamic acid to control the tumoral bleeding. After several days of care, the symptoms were resolved and the patient was discharged. Another follow up and workup diagnostic for the buccal mass was done after the symptoms resolved.

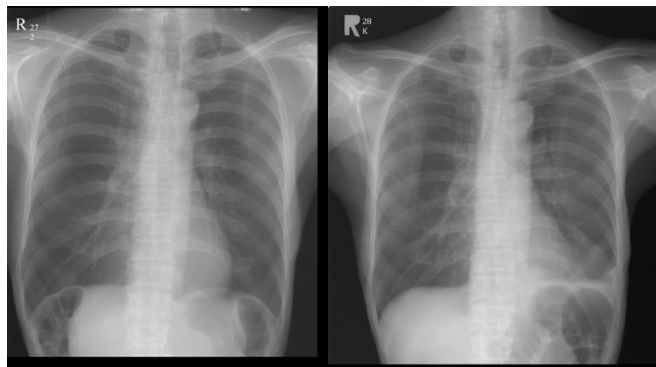


Figure 3: Plain chest radiograph of the 62-year-old man before and after conservative treatment.

Discussion

Chilaiditi syndrome was found in 0.25-0.28% of the population [3]. Actually, the chilaiditi syndrome was first described by Cantini in 1865, it was public after observation in the clinical examination in 1910 by Chilaiditi. Chilaiditi syndrome is a great mimicker with symptoms that imitate various medical and surgical emergencies [5]. Several theories have been proposed for chilaiditi syndrome, including the cause of anatomic distortions such as the laxity, absence, or elongation of the suspensory ligaments or the falciform ligament [6]. Some authors suggested that there is weak zone of the liver surface that created low resistance of minimal pressure, even pressure from fibrous bands between diaphragmatic muscle fibers [7]. Other causes were-

liver atrophy, elevated diaphragm due to phrenic nerve injury, development of sub diaphragmatic spaces after liver surgery, or obesity/weight loss that cause distortion of abdominal fat [3].

These diaphragmatic slips are often found on the right hemidiaphragm, with their concavity parallel to the falciform ligament [8]. The presence of diaphragmatic slips and associated hepatic grooves, in this case, created the 'indented' appearance of free air on the plain radiographs. A lung that is compressed by the distended loops of the bowel underneath his diaphragms will be the cause of his respiratory distress, similar to dyspnoea associated with ascites [3].

From table 1, we presented our literature search in the last 10 years for the study that reported Chilaiditi syndrome cases. All cases were found as an incidental finding from the Chest X-Ray and confirmed by abdominal CT Scan. Nearly all continents had reported at least one case of Chilaiditi syndrome, with a total of cases. Majority of the reported cases showed that the most common symptoms were epigastric pain with or without radiation to the right shoulder and constipation. Some cases were presented with shortness of breath. Not surprisingly, other authors reported that Chilaiditi syndrome could be mistakenly diagnosed as acute abdomen (peritonitis, ileus), lung disease, even myocardial infarction. Physical findings were not specific although most cases reported abdominal tenderness. All case reports reported 100% favorable outcome with conservative treatment. The symptoms resolved in the range of 6 hours to 2 months. Among cases that required surgery were those which coincidentally occurred with other abnormalities.

Table 1: Worldwide reported cases of Chilaiditi syndrome in the last 10 years (2009-2019) [5,10-54].

Case report	Country	Number of cases	Symptoms	Physical findings	Radiological findings	Management	Outcome
Malavade (2010)	India	1	Abdominal pain, dyspepsia, chest pain with angina-like symptoms	Sign of acute intestinal obstruction	X-Ray: elevated right hemidiaphragm ECG, Echocardiography, stress test: within normal limits	Conservative	Favorable
Okiro (2010)	Ireland	1	Abdominal pain, chronic constipation, respiratory distress	Unremarkable	X-Ray: elevated right hemidiaphragm	Conservative with laxative and enema	Favorable
Blevins (2011)	USA	1	Abdominal pain, vomiting	Unremarkable	Laparoscopy diagnostic	Conservative with laxative and dietary changes	Favorable
Nagem (2011)	Brazil	1	Epigastric pain, right shoulder pain	Tense and painful abdomen	X-Ray: air in subphrenic USG: no free fluid	Conservative with intravenous fluid	Favorable
Lin (2012)	Taiwan	6	Unspecified abdominal pain, constipation	Tenderness in the abdomen	X-Ray: air in subphrenic CT Scan	5 were managed conservatively with decompression and laxatives 1 was surgery due to coincidentally finding with volvulus	Favorable
Yin (2012)	USA	1	Abdominal pain	Sign of intestinal obstruction	CT Scan	Laparoscopic management due to failed conservative treatment	Favorable
Choi (2013)	Australia	1	Abdominal discomfort, nausea, vomiting	Unremarkable	Gastroscopy	Laparotomy due to coincidence with strangulated ileum	Favorable
Liu (2013)	Taiwan	1	RUQ pain	Abdominal distention, RUQ tenderness	X-Ray: elevated right hemidiaphragm CT Scan	Conservative with oxygenation, chest care, and pain control	Favorable
Chen (2014)	Taiwan	1	Chest pain	Unremarkable	X-Ray: increased intestinal gas CT Scan	Conservative for analgesic and intravenous fluid	Favorable
Ho (2014)	Taiwan	1	Epigastric pain	Epigastric distention	X-Ray: air in subphrenic	Conservative with decompression	Favorable

Mesquita (2014)	Israel	1	Abdominal pain, shortness of breath	Extreme abdominal distention, bloating and respiratory distress.	X-Ray: air in subphrenic USG: no free fluid	Conservation with decompression	Favorable
Weng (2014)	China	7	Abdominal tenderness, constipation	Abdominal pain	X-Ray: air in subphrenic	4 were managed conservatively with intravenous fluid and enema 3 were managed with laparotomy due to coincidence with rectal cancer, mesenteric lymphosarcoma, and atrophy of right liver lobe	Favorable
Zhou (2014)	China	1	RUQ pain	Tenderness at RUQ, decreased bowel sound	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Erdem (2015)	Turkey	2	Abdominal pain, shortness of breath, coughing	Unremarkable	X-Ray: elevated right hemidiaphragm	Conservative with laxative and high fiber diet. Respiratory tract infection was managed with antibiotics, oxygen, and analgesic	Favorable
Nayak (2015)	India	1	Pain at upper abdomen, constipation	Diffuse tendered abdomen	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Quiroga (2015)	Spain	1	Epigastric pain, constipation	Peritoneal irritation	X-Ray: elevated right hemidiaphragm CT Scan	Laparotomy (no other findings), finally diagnosed with Ogilvie syndrome	Favorable
Uygungul (2015)	Turkey	1	Epigastric pain	Epigastric tenderness	X-Ray: air in subphrenic	Conservative with pain management and intravenous fluid	Favorable
Watanabe (2015)	Japan	1	Upper abdominal pain, dyspnea	Peritoneal irritation	X-Ray: increased intestinal gas CT Scan	Laparotomy with coincidence finding of Bochdalek hernia	Favorable
Chen (2016)	USA	1	Epigastric pain, RUQ pain, nausea, vomiting	Tenderness in the epigastrium and RUQ	X-Ray: air in subphrenic CT Scan	Conservative with decompression	Favorable
Everngul (2016)	Turkey	1	Upper abdominal pain, constipation	Abdominal pain	X-Ray: air in subphrenic CT Scan	Conservative with pain management and intravenous fluid	Favorable
Garg (2016)	India	1	Abdominal pain, respiratory symptoms	Unremarkable	X-Ray: air in the subphrenic	Conservative	Favorable
Gurvits (2016)	USA	1	Bloating, constipation	Abdominal distention	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Ittayachen (2016)	India	1	Cough and fever	Decreased breath sound in the right infra-scapular area	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Jangouk (2014)	USA	1	Abdominal pain, constipation, bloating	Unremarkable	CT Scan	Conservative with intravenous fluid	Favorable
Takahashi (2016)	Japan	1	Right upper abdominal pain, vomiting	Right upper abdominal tenderness	X-Ray: air in subphrenic CT Scan	Laparotomy due to the development of ileus	Favorable
Zvezdin (2016)	Russia	1	Abdominal pain, respiratory distress	Unremarkable	X-Ray: elevated right hemidiaphragm	Conservative with decompression	Favorable
Aquilar-Garcia (2017)	Spain	1	RUQ abdominal pain, nausea, vomiting, bloating, constipation	Unremarkable	X-Ray: elevated right hemidiaphragm	Conservative with laxative and enema	Favorable
Cawich (2017)	Trinidad & Tobago	1	Upper abdomen pain	Tenderness at RUQ	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Garcia (2017)	USA	1	Abdominal pain, diarrhea	Abdominal distention	CT Scan	Robotic surgery laparotomy due to persistent abdominal pain	Favorable
Hountis (2017)	Greece	1	Abdominal pain, radiation to the shoulder	Abdominal tenderness	CT Scan MRI	Conservative	Favorable
Krzeminski (2017)	Poland	1	Epigastric pain radiating to the right side of the abdomen	Abdominal pain	X-Ray: air in subphrenic CT Scan	Conservative with decompression	Favorable

Laharwani (2017)	USA	1	N/A (the patient was intubated within the time of findings)	Abdominal distention	X-Ray: air in subphrenic	Conservation with decompression	Favorable
Naji-amrani (2017)	Morocco	1	Abdominal pain, cough with mucopurulent sputum	Bilateral bronchial rales	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Okiro (2017)	Japan	1	Abdominal and chest pain	Unremarkable	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Shinha (2017)	USA	1	Abdominal pain, nausea, vomiting	Diffuse tender abdomen, decrease bowel sound	X-Ray: air in subphrenic CT Scan	Conservative with decompression	Favorable
Taha (2017)	Oman	1	Epigastric and RUQ pain with radiation to right shoulder	No abnormalities	X-Ray: air in subphrenic USG: no free fluid CT Scan	Conservative with pain management and intravenous fluid	Favorable
Ali (2018)	UAE	1	Dyspnea, severe retrosternal chest pain	Pericardial rub, abdominal tenderness	X-Ray: elevated right hemidiaphragm CT Scan	Conservative	Favorable
Gad (2018)	Egypt	1	Abdominal pain, cough	Abdominal distention	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Harpain (2018)	Austria	1	Epigastric pain, nausea	Unremarkable	X-Ray: air in subphrenic	Conservative with decompression	Favorable
Hussain (2018)	UAE	1	Abdominal pain, cough	Abdominal distention, inspiratory, ronchi	X-Ray: elevated right hemidiaphragm	Conservative	Favorable
Kapania (2018)	USA	1	RUQ pain, severe cough	RUQ distention	CT Scan	Laparoscopic exploration due to long pain duration and frequency	Favorable
Karaman (2018)	Turkey	1	Epigastric pain, hiccups, constipation	Tenderness at the epigastric area and RUQ	X-Ray: air in subphrenic	Conservative with high fiber diet and laxatives	Favorable
Krishna (2018)	India	1	Respiratory distress, hemoptysis	Sign of liver cirrhosis	X-Ray: elevated right hemidiaphragm CT Scan	Conservative for chilaidditi and noninvasive ventilation, steroid, antibiotics for pneumonitis	Favorable
Luo (2018)	China	1	Abdominal pain constipation	Abdominal distention	X-Ray: air in subphrenic CT Scan	Conservative with decompression	Favorable
Ogasawara (2018)	Japan	1	Abdominal pain, constipation, respiratory distress	Abdominal pain	X-Ray: air in subphrenic	Conservative with a laxative (relieve the respiratory distress)	Favorable
Sunkara (2018)	USA	1	Abdominal pain	Abdominal pain	X-Ray: air in subphrenic	Laparotomy due to the presence of cecal perforation	Favorable
Vasileiadis (2018)	Greece	1	Epigastric and RUQ pain with radiation to right shoulder	Unremarkable	X-Ray: elevated right hemidiaphragm	Conservative	Favorable
Virmani (2018)	India	1	Abdominal pain (unspecified), shortness of breath	Tenderness of abdomen	X-Ray: air in subphrenic	Conservative with intravenous fluid and bronchodilators. Occur concomitantly with congenital diaphragmatic hernia	Favorable

The diagnosis of chilaidditi was only made with radiological evidence while in the absence of symptoms, the radiological finding just called chialiditi sign [55]. Criteria for Chilaidditi diagnosis were the elevation of right diaphragm above the liver by the intestine, the air in the subphrenic space, and depression of the superior margin of the liver below the left diaphragm [56]. The free air below diaphragm could lead to a conclusion of pneumoperitoneum if it were not confirmed by left lateral decubitus X-Ray which in Chilaidditi, the lucency did not shift upon the changing position [57]. Other imaging methods indicated for the differential diagnosis of Chilaidditi's syndrome include chest/abdominal computed tomography or MRI [58]. The trends from the analysis of the case was that, in pediatric patients, the workup diagnostics were made more carefully. Choi (2013) and Blevins (2011) even carried out laparoscopy and gastroscopy, respectively, to make sure that there were no other abnormalities that precipitate the abdominal pain [10,14].

Chilaidditi syndrome is managed conservatively with intravenous fluid, laxatives or enema, and nasogastric decompression. If it was complicated with intestinal obstruction, perforation, or strangulation, surgical intervention became mandatory. Patients with persistent recurring symptoms or failed to be managed conservatively were also managed surgically [59]. For the surgical approach, cecopexy was showed to be effective, unless gangrene or perforation necessitates bowel resection [60]. Although Saber et al. previously reported that 26% of patients require surgical management, the analysis of reported cases showed a lower number [4].

An interesting case reported by Quiroga et al. (2015) in Spain, which laparotomy was done for Chilaidditi syndrome in suspicious coincidence with peritonitis. The laparotomy showed no other abnormalities. Finally, the physicians revealed the use of psychotropic medications, which were then stopped, and the

patient recovered uneventfully. The clinicians made a diagnosis of Ogilvie syndrome, that happened in line with Chilaiditi syndrome [23].

Chilaiditi could be mistakenly diagnosed or found additionally with a variety of pulmonary and gastrointestinal disease. In this case, the author showed that Chilaiditi could be either presented with abdominal or respiratory symptoms. When evaluating asymptomatic patient, other abnormalities should be ruled out carefully [30]. The author recommended doing a CT Scan should be done to make sure the diagnosis of Chilaiditi syndrome. In the patient with persistent symptoms of the chilaiditi syndrome, attention needs to be given because this syndrome can be a precipitate factor for the additional condition of intestinal strangulation or obstruction. Therefore, a tight observation was needed to early diagnosis any sign of intestinal obstruction which resulted from the presence of Chilaiditi syndrome.

Conclusion

Clinicians should be more aware of the diagnosis of Chilaiditi syndrome with its various symptoms. Radiological evidence should be obtained for the confirmed diagnosis and conservative management was the modality choice.

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