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CASE REPORT



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Bilateral Chilaiditi Syndrome in a Nigerian Child with Severe Acute Malnutrition: A Case Report

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Abstract

Chilaiditi syndrome is characterised by abdominal pain, usually in the right upper quadrant, associated with a radiological finding of sub-diaphragmatic free air due to colonic interposition between the diaphragm and the liver. The Chilaiditi sign is the radiological finding of sub-diaphragmatic pneumoperitoneum due to colonic interposition between the diaphragm and the liver without any clinical symptom. It can be a source of diagnostic dilemma considering bowel perforation as a common differential leading to unnecessary exploratory laparotomy.

We report a four-year-old boy who presented with abdominal pain, fever, abdominal distension and diarrhoea with intermittent long-standing constipation. A plain abdominal radiograph in the erect position showed bilateral sub-diaphragmatic free air with associated haustration markings in free air. He was managed conservatively with antibiotics and nutritional rehabilitation with remarkable clinical improvement. While evaluating a child with an acute abdomen, the radiographic finding of free sub-diaphragmatic air, which is discordant with clinical signs, should heighten the suspicion of Chilaiditi syndrome. Knowledge of Chilaiditi syndrome is essential to avoid unnecessary surgical intervention while considering bowel perforation as a common cause of sub-diaphragmatic air.

Keywords: Abdominal pain, Chilaiditi syndrome, Chilaiditi sign, Diarrhoea, Sub-diaphragmatic, Pneumoperitoneum.

Introduction

Chilaiditi syndrome is characterised by symptoms such as abdominal pain associated with radiological findings of colonic interposition between the diaphragm and the liver, which is seen as sub-diaphragmatic free air. Chilaiditi sign is the radiological finding of sub-diaphragmatic pneumoperitoneum due to colonic interposition between the diaphragm and the liver. Chilaiditi syndrome is a clinical presentation characterised by a Chilaiditi sign on a radiograph. We report a rare case of a four-year-old Nigerian child with severe acute malnutrition who presented on account of abdominal pain and was subsequently found to

have a bilateral Chilaiditi sign on erect view of abdominal X-ray.

Case presentation

A 4-year-old boy presented to the Emergency Paediatric Unit of the Federal Medical Centre, Nguru, Nigeria, in June 2023, on account of abdominal distension, abdominal pain and fever of one-week duration and diarrhoea of five days duration. The abdominal pain reportedly started as periumbilical but later involved the upper abdomen, with no known aggravating or relieving factor. The pain was severe enough to have disturbed his sleep and oral feeding. The diarrhoea was mucoid with a

frequency of three to five times a day but nonbloody. There was no vomiting, jaundice or urinary symptoms. However, there was a history of intermittent constipation which usually subsided after defaecation.

On examination, he was pale, anicteric, not cyanosed but febrile with a temperature of 38.3°C. He was wasted with a body weight of 7.8kg and a height of 96cm (48.8% of expected weight and 95% of expected height), respectively. His weight-for-height Z-score was < -3SD on the weight-for-height chart, and the mid-arm circumference was 10cm. He was not dyspnoeic and had a respiratory rate of 22 cycles/minute with vesicular breath sounds and no crepitations. He was saturating at SpO₂ 99% in room air. He had normal volume pulses, which were regular and synchronous. The pulse rate was 102/minute and the first and second heart sounds were normal without cardiac murmur. Abdominal examinations revealed a distended abdomen with vague tenderness over the abdomen without guarding and no palpable organomegaly. Bowel sounds were hypoactive. He was conscious and alert, and the muscle tone was reduced across the limbs. The initial diagnosis was perforated viscus secondary to enteric fever with background severe acute malnutrition to keep in view hypokalaemia. He was placed on nil per os, a nasogastric tube was inserted for gastric decompression, and a catheter was also used for urinary output monitoring. Intravenous ceftriaxone. gentamycin and metronidazole commenced. He also had an intravenous infusion of 4.3% Dextrose in 1/5th Saline made up to 8.4% dextrose concentration at a maintenance rate. The surgical team reviewed the child and considered possible exploratory laparotomy.

The haemogram showed packed cell volume of 22.2%, leucocytosis with white cell count of 18, 900/mm³ (differential count: neutrophil 71.8%, lymphocytes 18.9% and monocytes 8.8%). The serum electrolytes, urea and creatinine revealed hypokalaemia of 1.9mmol/l, hyponatraemia of

128mmol/l, and hypochloraemia of 85mmol/l, while other parameters were normal. Urinalysis and random blood glucose (RBG) were also within normal limits. Blood film microscopy was negative for malaria parasites. Stool Gene Xpert did not detect Mycobacterium tuberculosis. The deranged serum electrolytes were corrected via intravenous fluid infusions and a repeat serum E/U/Cr about three days after admission revealed a normal serum potassium level (3.4mmol/l).

Abdominal ultrasonography revealed that the visualised distal part of the large bowel was loaded with faecal matter with poor peristaltic activity. No ascites or gases were noted. The liver, gallbladder, spleen, paracolic areas, urinary bladder, and kidneys were grossly normal. However, an erect view of the plain abdominal radiograph revealed free air under the hemidiaphragm bilaterally, with colonic faecal loading (See Figure 1). Hence, a diagnosis of Chilaiditi was made.

Following a review of the radiograph and the resolution of abdominal pain on the third day of admission, the child was recommenced on graded oral feeding and was transferred to a stabilisation centre for continued in-patient management as a case of a severely acutely malnourished child. An abdominal CT scan could not be done because the facilities were unavailable. All the admitting symptoms were resolved by the fifth day of admission. He was discharged to an outpatient therapeutic programme (OTP) after six days of admission. He is currently being followed up in the clinic. The last clinic visit was two months after discharge.



Figure 1: Abdominal radiograph (erect view) showing bilateral sub-diaphragmatic free air (Arrow) - Chilaiditi sign.

Discussion

Radiological sub-diaphragmatic free air in a child with an acute abdomen is a surgical emergency and is most commonly due to bowel perforation.¹ Sub-diaphragmatic free air occurs in more than 90% of patients with bowel perforation.² Other possible causes are peritoneal dialysis, post-operative free intraperitoneal gas, gallstone ileus, biliary enteric fistula, post-adenotonsillectomy, subdiaphragmatic abscess, sub-pulmonary pneumomediastinum, gas-containing haematoma and Chilaiditi sign or syndrome. 1,3,4 Peritoneal dialysis and post-operative state result in sub-diaphragmatic free air from air entrance into the peritoneum during the procedure. In gallstone and biliary enteric fistula, the sub-diaphragmatic air arises from the air in the gall bladder getting into the peritoneum. In sub-diaphragmatic abscess, subdiaphragmatic free air may be caused by gasproducing bacteria such as Clostridium welchii. Chilaiditi sign is seen as the sub-diaphragmatic free gas and haustration within the gas. This is referred to as Chilaiditi syndrome when there are symptoms such as abdominal pain and constipation in the presence of Chilaiditi signs. Chilaiditi syndrome should be suspected if there is radiological sub-diaphragmatic free air

and discordant clinical features. Although the first description of the interposition of the colon between the liver and the right hemidiaphragm was published by Cantini in 1865, the Chilaiditi sign was named after Demetrius Chilaiditi, a Greek Radiologist in 1910 after he reported three cases with radiological evidence of colonic interposition between the diaphragm and the liver while working in Vienna.^{5,6} The Chilaiditi sign is most commonly seen in older people with an incidence of ~ 1% worldwide and has a male predilection of 4:1.7. It has a global incidence of 0.025–0.28% and a poorly understood aetiology.⁷ It is more common over the right side than the left, 8 but it may also be bilateral.^{9,10} An extensive review of the literature found only 32 paediatric cases of Chilaiditi syndrome, but the index case might be the first reported case in a Nigerian, especially in a child. The index child had bilateral Chilaiditi syndrome probably because of co-existing severe acute malnutrition [defined by MAC of 10cm (<11.5cm) and weight for height Z-score < -3 SD] that could lead to partial loss of tone of the falciform, phrenicocolic and splenophrenic ligaments as a result of protein deficiency. This poor ligamental supports might predispose to colonic malpositioning on both sides of the diaphragm. Nutritional rehabilitation might have strengthened the ligaments, thus repositioning the colon.

Whilst some individuals are asymptomatic and the Chilaiditi sign is observed incidentally on a radiograph (chest or erect view of the abdomen),¹¹ some may present with varying symptoms such as abdominal discomfort, abdominal pain, nausea. vomiting. constipation, flatulence, respiratory distress, chest pain, difficulty with breathing, features of intestinal obstruction as well as features of intestinal perforation.^{8, 12-14} These symptoms usually arise from the pressure effect of colon interposition between the diaphragm and the liver. The diagnosis is usually clinicoradiological and due to its rarity, non-specific presentation, and intermittency in occurrence, it

may serve as a potential mislead. Imaging modalities that can assist in the diagnosis are chest radiography or erect view of a plain abdominal radiography, abdominal ultrasound scan or computerised tomography (CT) scan. Due to its high sensitivity, a CT scan may be indicated for confirmation of the subdiaphragmatic free air in doubtful circumstances.

The mainstay of treatment is usually conservative; these include dietary modification and laxatives depending on the presenting symptoms. Surgical rectification in colopexy or resection might be rarely needed, especially if associated with volvulus, intestinal ischaemia and perforation. ¹⁵

Evaluation of a child with abdominal pain may require imaging; if there is radiological sub-diaphragmatic free air and discordant clinical features, the possibility of Chilaiditi syndrome is heightened. When available, further confirmation with a CT scan can be done to avoid unnecessary surgical exploration.

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