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Case Report

Left-sided acute appendicitis in a patient with situs inversus totalis: A case report and a comprehensive review ☆,☆☆

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ABSTRACT

AA is a frequent surgical condition that demands urgent intervention. It accounts for approximately 6% of all emergency department visits. Situs inversus is a rare condition in which the orientation of asymmetric organs is a mirror image of normal anatomy. It can be partial (involving either the abdominal or thoracic cavities) or complete (situs inversus totalis: transposition of both abdominal and thoracic organs). SIT is very rare, with an incidence of 1 per 5000 to 10,000 live births. It is inherited in an autosomal recessive pattern with incomplete penetrance. LSAA is very rare and can happen in association with other congenital abnormalities such as situs inversus, midgut malrotation (MM), or a usually long right-sided appendix projecting into the left lower quadrant. SIT is responsible for greater than 67% of left-sided appendicitis cases. Due to atypical clinical presentation, the diagnosis of AA can be difficult and often delayed. Hence, a complete medical history, physical examination, laboratory tests, and imaging tools are necessary to reach a correct diagnosis in a timely manner and prevent complications like abscesses, perforations, and peritonitis.

We report a case of a 50-year-old male with symptoms of left lower abdominal pain along with fever, nausea, vomiting, and loose stools that were later diagnosed as LSAA in the setting of SIT.

Abbreviations: LSAA, Left-Sided Acute Appendicitis; SIT, Situs Inversus Totalis; AA, Acute Appendicitis; MM, Midgut Malrotation; US, Ultrasound; CT, Computed Tomography.

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Introduction

AA is a frequent surgical condition that demands urgent intervention [1]. It accounts for approximately 6% of all emergency department visits [2]. According to the Global Burden of Disease Study 2019, the age-standardized incidence of AA was 229.9 per 100,000 individuals [3]. Situs inversus is a rare condition in which the orientation of asymmetric organs is a mirror image of normal anatomy. It can be partial (involving either the abdominal or thoracic cavities) or complete (situs inversus totalis: transposition of both abdominal and thoracic organs) [4]. SIT is very rare, with an incidence of 1 per 5000 to 10,000 live births. It is inherited in an autosomal recessive pattern with incomplete penetrance [5]. It can manifest as an individual entity or might be linked with conditions such as primary ciliary dyskinesia or Kartagener's syndrome [6]. LSAA is very rare and can happen in association with other congenital abnormalities such as situs inversus, midgut malrotation (MM), or an unusually long right-sided appendix projecting into the left lower quadrant [7]. SIT is responsible for greater than 67% of left-sided appendicitis cases [8]. Due to atypical clinical presentation, the diagnosis of AA can be difficult and often delayed [4]. Hence, a complete medical history, physical examination, laboratory tests, and imaging tools are necessary to reach a correct diagnosis in a timely manner and prevent complications like abscesses, perforations, and peritonitis [9].

We report a case of a 50-year-old male with symptoms of left lower abdominal pain along with fever, nausea, vomiting, and loose stools that were later diagnosed as LSAA in the setting of SIT.

Case Presentation

We present the case of a 50-year-old male who arrived at our center with a chief complaint of left lower abdominal pain persisting for 7 days. He had experienced episodes of nausea, vomiting, loose stools, and a raised temperature of up to 100 degrees Fahrenheit at home. He had no urinary symptoms. The patient had been prescribed antispasmodics, urine alkalizer, and ciprofloxacin by a pharmacist at a local polyclinic who suspected renal or ureteric colic. Despite the initial treatment, the patient's pain and symptoms worsened over the course of 7 days, prompting his visit to our facility.

The patient had no significant past medical or surgical history. However, he mentioned an encounter with a doctor in a remote village who had told him that his heart was located on the opposite side during a tuberculosis (TB) screening health camp. On physical examination, the patient exhibited moderate tenderness in the left iliac fossa without rebound tenderness or guarding. His temperature was 102 degrees Fahrenheit. No pain was elicited at McBurney's point. Laboratory investigations showed a total white blood cell count of 14,000 cells/cubic millimeter with 82% neutrophil predominance. Amylase and lipase levels were within normal limits.

The patient was initiated on a nil-per-oral (NPO) diet and underwent initial pain management and fluid resuscitation. Subsequently, a CT abdomen and pelvis without contrast was performed due to resource constraints and the emergent nature of the situation. The CT images, obtained from a third-generation CT machine with a 5 mm slice thickness, were reconstructed into multi-planar images.

The CT revealed dextrocardia along with an ascending aorta to the left, a descending aorta to the right, and the pulmonary trunk to the right side of the chest. In the abdomen, the liver was situated on the left side, while the stomach and a single spleen were positioned on the right. The gastrointestinal tract exhibited complete mirror imaging, with the stomach, duodenojejunal flexure, and descending colon on the right, and the ileocaecal junction, caecum, and ascending colon on the left (Fig. 1). A distended, blind tubular structure measuring 13 mm in diameter was identified in the left lower quadrant, representing the distended appendix (Fig. 2). Mild peripheral fat stranding was also appreciated along with the distended appendix on the left side (Fig. 3). A portable Ultrasound right after the CT revealed a dilated, aperistaltic, noncompressible, and inflamed appendix corroborating the CT findings (Fig. 4).

Based on the diagnosis of acute appendicitis with situs inversus totalis, the patient was promptly referred to a tertiary center equipped with surgical services. Urgent laparoscopic appendectomy under general anesthesia was performed. De-

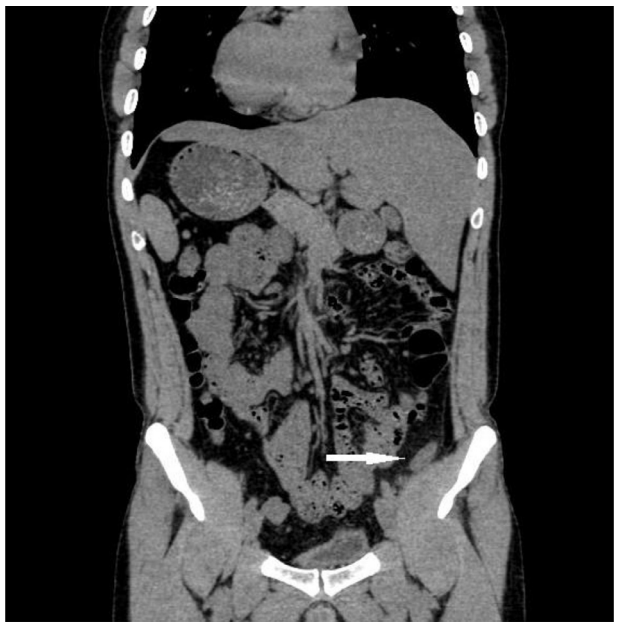

Fig. 1 – Noncontrast CT (coronal view) revealing dextrocardia with liver positioned on the left side and the spleen and the stomach positioned on the right side. The duodenojejunal flexure and the descending colon are positioned on the right while the ileo-caecal junction, caecum and ascending colon are positioned on the left side. A distended, blind tubular structure measuring 13.4 mm in diameter is identified in the left lower quadrant, representing the distended appendix (shown by the white arrow).

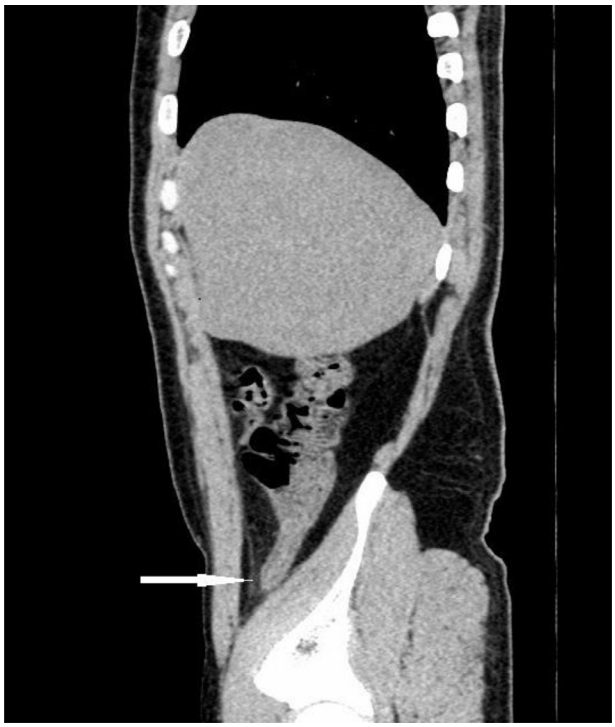

Fig. 2 – Noncontrast CT (sagittal view) showing a distended, blind tubular structure, representing a distended appendix (shown by the white arrow).

spite the challenging anatomical variation, intraoperative observations revealed a distended and inflamed appendix with periappendicular fluid collection and mild periappendiceal fat stranding (Fig. 5). The specimen (Fig. 6) was sent for biopsy. The patient received pain medication and intravenous antibiotics (Metronidazole and Amoxicillin + Clavulanic acid). He was discharged on the fourth postoperative day after meticulous wound assessment and counseling regarding his rare condition. The histopathological report showed findings of appendicitis. A 1-week follow-up showed favorable progress, and the patient resumed his normal activities within a month without any signs of postoperative complications.

Discussion

Situs inversus is an uncommon condition where the orientation of asymmetric organs mirrors the normal anatomy. It is a rare autosomal recessive congenital anomaly that occurs when there is a 270-degree rotation clockwise, resulting in the complete transposition of all intra-abdominal organs [10]. The reported incidence of SIT in the literature is 0.001% to 0.01% in the general population [7]. Acute appendicitis (AA) is the most common surgical emergency and usually manifests as right iliac fossa pain. However, due to significant variations in the location of the vermiform appendix, about one-third of patients with AA have abdominal pain in sites other than the right iliac fossa [11]. Left lower quadrant abdominal pain as a presentation of acute appendicitis is usually uncommon [7]. The incidence of acute appendicitis in the case of SIT, as reported in the literature is 0.016% to 0.024% [7]. LSAA occurs between the ages of 8 and 63 years and is 1.5-fold more frequent in men than in women [12]. LSAA is very rare and can happen in association with other congenital abnormalities such as SIT and midgut malrotation [7], with SIT being responsible for more than 67% of the cases [8]. Our case describes a 50-year-old male with complaints of left lower abdomen pain along with vomiting, fever, and loose stools. LSAA may also be an unusual presenting feature of a long right-sided appendix extending into the left lower quadrant or of a mobile and redundant caecum [11,13]. The differential diagnosis of left lower quadrant includes bowel obstruction, acute sigmoid diverticulitis, strangulated or incarcerated hernia, small bowel enteritis, Meckel's diverticulum, ruptured ovarian cyst or ectopic pregnancy, ureteric colic, acute epididymitis, psoas abscess, and acute appendicitis [2]. Diagnosing LSAA in SIT can be challenging because, despite abdominal viscera displacement, the pain from left-sided acute appendicitis in situs inversus can be localized to the right lower quadrant [4]. MM is another congenital anomaly that serves as a differential for LSAA. MM occurs due to either nonrotation or incomplete rotation of the primitive intestinal loop around the axis of the superior mesenteric artery during fetal development. Its reported incidence in the literature varies from 0.03% to 0.5% in the live births [7]. Usually, MM presents in the first month

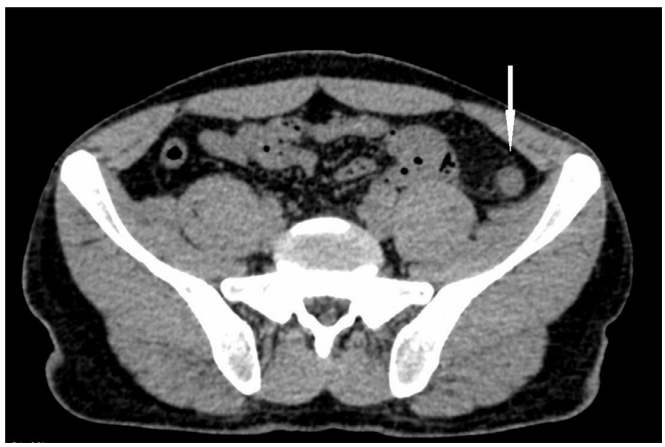

Fig. 3 – Noncontrast CT (axial view) showing a distended appendix with mild peripheral fat stranding noted on the left side.

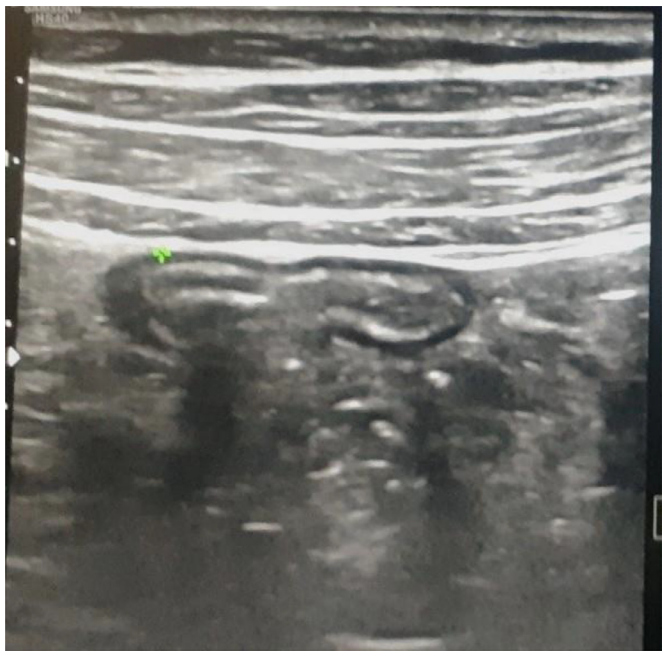

Fig. 4 – Ultrasound of the abdomen and pelvis revealing a dilated, aperistaltic, noncompressible, and inflamed appendix.

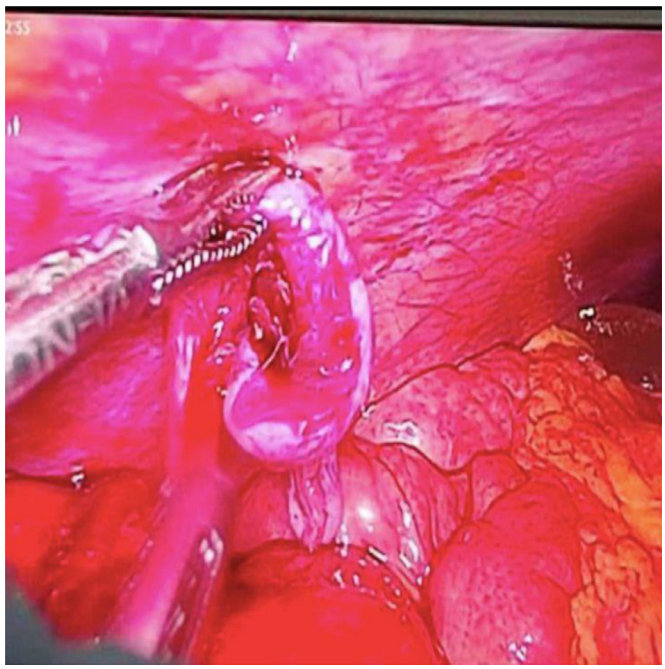

Fig. 5 – Laparoscopic image of an acutely inflamed appendix.

of life with bilious vomiting and is rarely seen in adults [7]. Diagnosis of LSAA in these patients with SIT can be made by means of physical examination, chest X-ray, ECG, USG, CT scan, and laparoscopy [10,11]. Physical examination may reveal right-sided heart sounds, left lower quadrant tenderness, palpable liver edge on the left side, and the right testicle lower than the left [7]. X-rays though not usually applied for diagnosing SIT and LSAA, but the presence of dextrocardia and right-sided gastric bubble may provide a valuable clue towards the diagnosis [4]. Though there is no evidence of any X-ray findings in our patient, he mentioned an encounter with a doctor in a remote village who had told him that his heart was located on the opposite side during a TB screening health camp. US also serves as a useful modality for diagnosing AA but it is operator-dependent [4]. CT scan with a reported sensitivity of 90% to 98%, serves as a valuable tool in diagnosing AA in

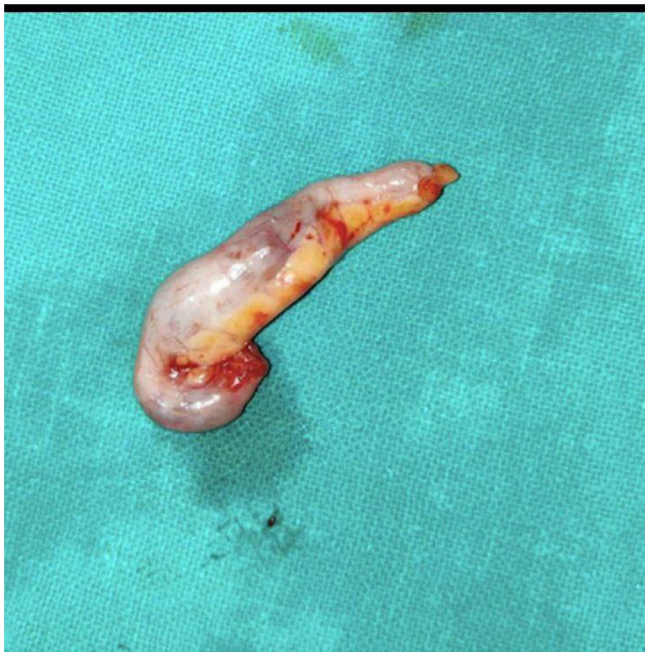

Fig. 6 – Resected appendix after laparoscopic appendectomy which shows inflammatory changes.

patients with SIT, because of its ability to outline the abnormal location of the appendix [11]. The main indicators of AA in CT scans include peri-appendiceal inflammatory changes, an appendix with a diameter of more than 6 mm, or a wall thickness of more than 3 mm [14,15]. CT scan and US were also done in our case and the findings were consistent with that of the literature. Although laparoscopic appendectomy is challenging in these cases, it is still the standard approach for management [1]. The advantages of this technique are rapid postoperative recovery, shorter hospital stay, less surgical stress, and lower postoperative complications [16]. Moreover, laparoscopic appendectomy is of great utility in situations where clinical and radiological findings are uncertain or when the appendix is positioned atypically, as it eliminates the need for extensive incisions to guarantee sufficient access [17]. Nonetheless, there is not a predefined trocar insertion site for these specific cases, and the surgeon should adjust port placement according to laparoscopic surgical principles [18]. Our case was managed via laparoscopic appendectomy and the port placement was done according to the comfort of the surgeon (left hand as the working hand for right-handed surgeon).

Conclusion

SIT with LSAA must be kept in the differential diagnosis of a patient, if he/she presents with acute onset left lower quadrant pain, apart from the usual differentials pertaining to the normal location of the visceral organs. Complete medical history, physical examination, laboratory tests, and imaging tools (X-ray, US, CT scan) are necessary to reach a correct diagnosis in a timely manner and prevent complications like abscesses, perforations, and peritonitis. In resource-limited countries like Nepal, where electronic medical records are not feasible, it's essential to thoroughly document early-detected congenital anomalies. This documentation aids in forming accurate medical and surgical diagnoses, considering past malformations.

Ethical approval

The study is exempt from ethical approval in our institution.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Role of generative AI

None.

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