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Acute Abdomen in Situs Inversus Totalis

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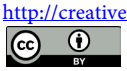


Abstract

Introduction: Situs Inversus (SI) is a rare autosomal recessive or an X-linked condition resulting in an abnormal mirror image abdominal organs position with a rate of 0.01% among the population. When SIT individuals present clinically with abdominal pain, the diagnosis can be challenging to make without any advanced imaging modality, as the symptoms are often nonspecific and do not align with the normal anatomical positioning of the abdominal organs, and this could cause a delay in diagnosis and management. In this paper, we discuss the challenging pathway in diagnosing and managing surgical abdominal emergencies in patients with situs inversus. **Case Presentation:** We present a 22-year-old male medically free who came to the emergency room with abdominal pain, vomiting, and constipation following an appendectomy. The patient had undergone laparoscopic appendectomy in another hospital for perforated appendicitis almost one month ago, during the postoperative period, an abdominal drain was kept for a few days until discharge. Computed tomography (CT) was done and showed a case of situs inversus totalis (SIT) with a completely mirrored image of the normal abdomen orientation. The decision was made to admit the patient for a trial of conservative management as treatment for adhesive bowel obstruction. Patient was kept nil per os (NPO) and on intravenous fluids (IVF), with electrolyte correction and frequent physical examination reassessment. A gastrografffine challenge test was done. Patient ingested around 100 ml of hyperosmolar contrast mixed with water. During re assessment patient had passed bowel motion due to the laxative effect of gasrtograffine and was stable and fit for discharge. Since it is known that patients with SIT might have an undiagnosed congenital heart disease, cardiology consultation was sent, and ECHO (Echocardiography) was arranged. ECHO revealed ejection fraction (EF) of 50%, reduced left ventricular (LV) function, tricuspid regulation, and mitral regurgitation. Patient was started on Bisoprolol 1.25 mg, Lisinopril 2.5 mg, and Dapagliflozin 10 mg for cardiomyopathy. **Discussion:** SIT is a rare clinical finding in the day-to-day practice however when presented, diagnosis and surgical approach can be challenging.

Our aim in this paper is to discuss proper examination of patients with abdominal pain and SIT, the need for cardiac screening in SIT, and surgical approaches in acute abdomen and SIT. The location of the pain in acute appendicitis with anomalies varies, but almost 70% of patients had left lower quadrant pain, other presented with right lower quadrant pain, peri-umbilical, and diffuse abdominal pain. Identifying anomalies during acute settings can be difficult, in one study; preoperative diagnosis was made in 83.6% of cases based on radiological findings, while 16.4% were identified during surgery. The diagnosis of acute appendicitis in such cases can often be complex, and delay in surgical management may contribute to increased morbidity and mortality. Misdiagnosis is more prevalent in cases where patients present with atypical symptoms, such as pain localized to unexpected areas. The occurrence of acute appendicitis in conjunction with situs inversus totalis (SIT) or mirror image malformation (MM) is uncommon, with an incidence ranging from about 0.016% to 0.024%. Regarding the appropriate surgical technique in dealing with patients diagnosed with anatomic anomalies. Laparoscopic exploration is a useful tool when clinical and radiological findings are inconclusive, especially when the appendix is in an unusual anatomical location. This method eliminates the need for large incisions and provides easier access for the trained surgeon. Laparoscopy allows for a thorough examination of the entire abdominal cavity, helping to confirm the initial diagnosis and identify any other pathological conditions. There is no fixed protocol for trocar placement in these unusual cases, and the surgeon should adjust port positioning based on key laparoscopic principles, such as triangulation and ergonomics. **Conclusion:** We suggest detailed history taking and examination in patients with acute abdomen and unclear diagnosis. Always remember to keep a broad differential diagnosis in mind. We recommend screening for any cardiac anomalies in patients with SIT since congenital heart disease is observed in 3% to 5% of these cases. And lastly, to not hesitate with liberal use of laparoscopic exploration as a diagnostic tool in unclear cases of abdominal pain.

Keywords

Situs Inversus, Situs Inversus Totalis, Acute Appendicitis, Bowel Obstruction

1. Introduction

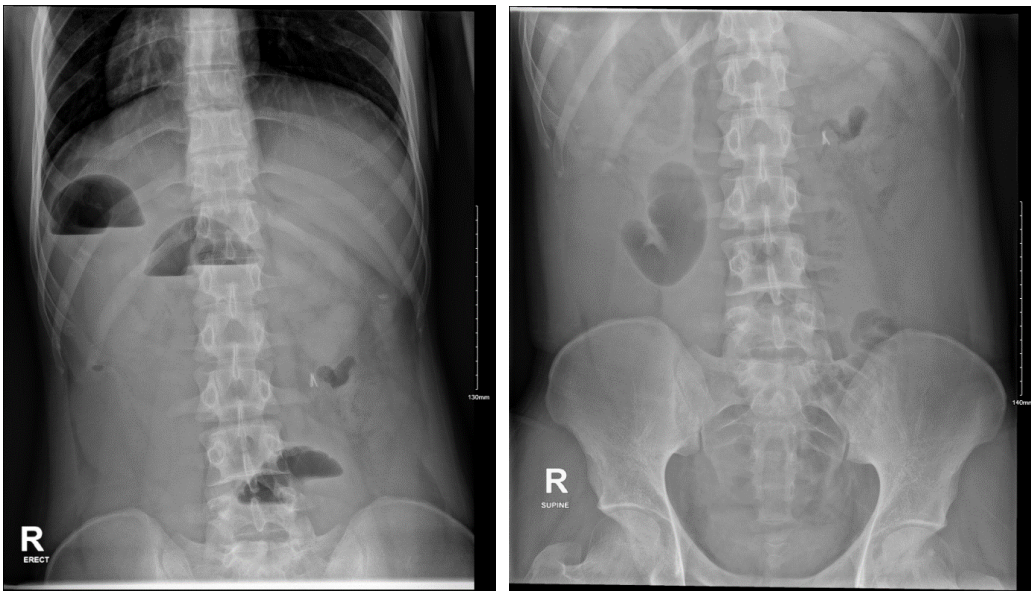
Situs Inversus (SI), Situs Oppositus, or Situs Viscerus Inversus is a rare autosomal recessive or an X-linked condition resulting in an abnormal mirror image abdominal organs position with a rate of 0.01% among the population [1]. The coexistence of abdominal situs inversus and abnormal heart position known as dextrocardia is termed situs inversus totalis (SIT), and it has an incidence of approximately 1 in 200,000 [2]. Congenital heart disease is associated with SIT in 3% - 5% of patients [3]. Often SIT can go undiscovered for many years because it is a non-pathological variant of the normal anatomy, but it is linked to some clinically significant conditions, such as primary ciliary dyskinesia affecting the lungs and

mucus secreting membranes throughout the body. Further, the combination of situs inversus, chronic sinusitis, and bronchiectasis is known as Kartagener syndrome. The pathogenesis of SIT is unknown, however speculations of genetic and molecular disturbance to normal anatomic rotation during embryonic periods is the main hypothesis [2]. When SIT individuals present clinically with abdominal pain in particular, the diagnosis can be challenging to make without any advanced imaging modality, as the symptoms are often nonspecific and do not align with the normal anatomical positioning of the abdominal organs, and this causes a delay in diagnosis and management. For example, if a patient presents with lower abdominal pain, acute appendicitis would be suspected as top differential diagnosis, however in cases of SIT the appendix can be found elsewhere and other diagnoses must be considered. Another important surgical presentation in SIT is volvulus; which is a torsion of one part on the intestine, since these patients at higher risk for gastrointestinal malrotations accounting for 40% [1]. In this paper, we discuss the challenging pathway in diagnosing and managing surgical abdominal emergencies in patients with situs inversus.

2. Case Presentation

We present a 22-year-old male previously medically free who came to the emergency room with abdominal pain, vomiting, and constipation following an appendectomy a month prior in another hospital. The pain was on-off and localized to the lower abdomen, it started two weeks after the surgery, with progressive worsening. The pain was associated with multiple episodes of bilious vomiting related to meals and no bowel motion for one day, as it is not his usual bowel habits. The patient had undergone laparoscopic appendectomy in another hospital for perforated appendicitis almost one month ago, during the postoperative period, an abdominal drain was kept for a few days until discharge, it was an uneventful hospitalization. On initial examination, the patient was vitally stable, abdominal exam revealed previous laparoscopic ports in the suprapubic, periumbilical and left lower quadrant regions (as the usual ports location in laparoscopic appendectomy), upon auscultation there were increased bowel sounds, and mild lower abdominal tenderness but no rigidity Laboratory works were also unremarkable with no leukocytosis, normal hemoglobin, and only mildly elevated C-Reactive Protein (CRP = 22.4). Abdominal X-ray (AXR) was initially performed and showed multiple air fluid levels on erect pictures (**Figure 1**). We preceded with abdominal computed tomography (CT) with intravenous (IV) as shown in (**Figure 2**). CT reported a case of situs inversus totalis (SIT) with a completely mirrored image of the normal abdomen orientation. The aorta was on the right side. He had small bowel obstruction with multiple dilated bowel loops with fluid and air-fluid levels reaching maximum diameter of 4.1 cm with an adhesive band at the transition zone. There is also placement of the entire large colon to the left side which appear completely collapsed. Evidence of surgical clips are seen in the left upper quadrant likely related to the prior surgery. Evidence of polysplenia

with multiple splenules noted as multiple round homogenous hyperdense enhancing structures in the central small bowel mesentery. Finally, the liver is noted in the left side. According to the patient, he knows about his anomaly as told by his parents when he was a kid but never needed to seek medical attention in his life prior to his acute appendicitis presentation.



(a) (b)

Figure 1. Initial AXR. (a) Upfront abdominal X-ray, showing multiple air fluid levels; (b) Supine abdominal X-ray, showing intraluminal air in gith upper quadrant.

Figure 2. Initial CT.

The decision was made to admit the patient for a trial of conservative management as treatment for adhesive bowel obstruction. The patient was kept nil per os (NPO) and on intravenous fluids (IVF), with electrolyte correction and frequent physical examination reassessment. There was no indication of nasogastric tube (NGT) as there were no repeated episodes of vomiting and no significantly dilated stomach. On days 1 - 5, the patient had no episodes of vomiting thus progression of diet started gradually.

On days 6 - 7, since the patient has not had bowel motion for over 1 week. A gastrograffine challenge test was done. Patient ingested around 100 ml of hyperosmolar contrast mixed with water. AXR was done 4 hours after (Figure 3) and showed smooth passage of oral contrast pass the transition zone. During re assessment patient had passed bowel motion due to the laxative effect of gasrtograffine and was stable and fit for discharge.

Since it is known that patients with SIT might have an undiagnosed congenital heart disease, cardiology consultation was sent, and ECHO (Echocardiography) was arranged. ECHO revealed ejection fraction (EF) of 50%, reduced left ventricular (LV) function, tricuspid regulation, and mitral regurgitation. Patient was started on Bisoprolol 1.25 mg, Lisinopril 2.5 mg, and Dapagliflozin 10 mg for cardiomyopathy. Outpatients' appointments with cardiology were given.

Unfortunately, the patient did not attend his surgery clinic follow-ups.

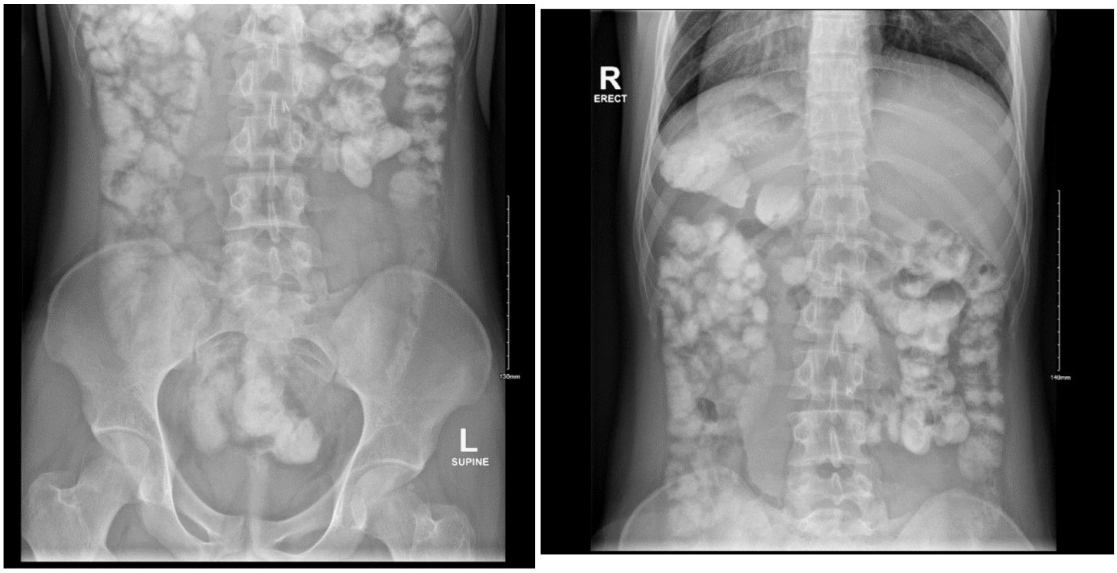


Figure 3. Pre discharge AXR.

3. Discussion

SIT is a rare clinical finding in the day-to-day practice however when presented, diagnosis and surgical approach can be challenging. Our aim in this paper is to discuss proper examination of patients with abdominal pain and SIT, the need for cardiac screening in SIT, and surgical approaches in acute abdomen and SIT.

Late advancement in fetal screening programs has led to earlier anomalies de-

tection in utero. In a systematic review of approximately seventy cases, they included cases of anatomic anomalies and acute appendicitis, 46 cases had SIT, while other anomalies included Kartagener's syndrome and midgut malrotation. Further, 84% of all cases had unknown anomalies, and only 16% were known due to earlier screening or radiological exam [4].

As mentioned earlier, congenital heart disease is associated with situs inversus and dextrocardia in 3% - 5% of cases. This reflects the importance of further cardiac work up for any patient with SIT and dextrocardia. We recommend through cardiac assessment especially as part of the pre operative anesthesiology clearance for those patients. Our patient will need regular follow up and long term medications for his cardiomyopathy, also strenuous excessive and land military duties will be somehow restricted which will affect his job and quality of life.

The diagnosis of acute appendicitis in our case was made in the previous hospital clinically (based on history and examination). Since patient presented late with perforated appendix and pain was not specific to one location. They used the usual ports placement in the paraumbilical, suprapubic and left abdominal. After that, they were faced with different picture of acute appendicitis than the usual right sided appendix. Examination of patient with SIT is not different from abdominal examination of the general population. Starting with vital signs including heart rate, blood pressure, and temperature. Then the general appearance of the patient whether he looks pale, sick or in pain. During local abdominal examination start with inspection for any deformity, ecchymosis, previous scars and start with palpation for tenderness, feeling for any masses, rigidity, etc. Palpation at McBurney point (2/3 distance between anterior superior iliac spine and umbilicus) is a specific hallmark for acute appendicitis. Other tests including Dunphy's cough test, Iliopsoas and Obturator signs can be performed for acute appendicitis and can yield a positive test based on the location of the appendix. Then complete the exam by percussion for any ascites, and auscultation for bowel sounds. Listing a differential diagnosis for abdominal pain is made mainly on the location of pain and the corresponding organ. As mentioned earlier the top diagnosis for right lower abdominal pain is acute appendicitis. The location of the pain in acute appendicitis with anomalies varies, but almost 70% of patients had left lower quadrant pain, other presented with right lower quadrant pain, peri-umbilical, and diffuse abdominal pain [4]. In (**Figure 4**) is a comparison of normal pathology based on underlying organ placement in normal population versus people with SIT.

Identifying anomalies during acute settings can be difficult, in one study; preoperative diagnosis was made in 83.6% of cases based on radiological findings, while 16.4% were identified during surgery [4]. The diagnosis of acute appendicitis in such cases can often be complex, and delay in surgical management may contribute to increased morbidity and mortality. Misdiagnosis is more prevalent in cases where patients present with atypical symptoms, such as pain localized to unexpected areas, such as in cases with abnormal anatomy [4]. The occurrence of acute appendicitis in conjunction with situs inversus totalis (SIT) or mirror image

malformation (MM) is uncommon, with an incidence ranging from about 0.016% to 0.024% [5].

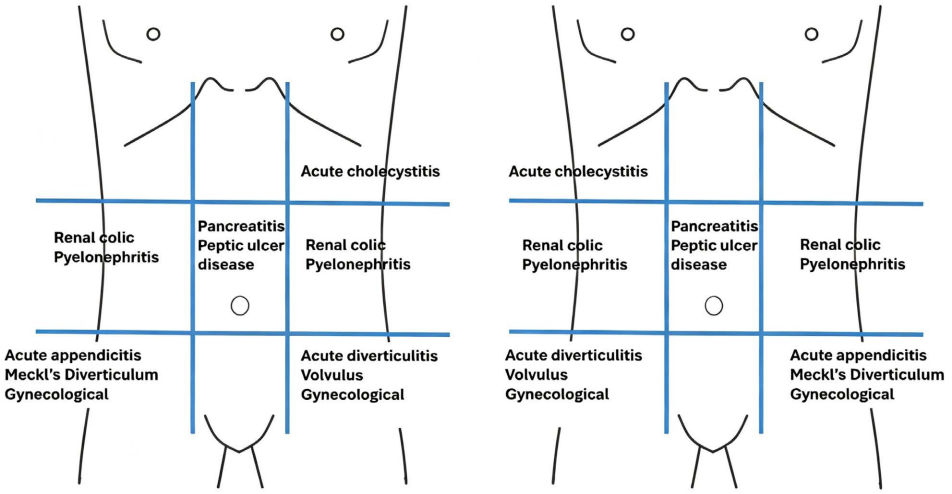


Figure 4. Abdominal pain and corresponding pathology among general population versus SIT.

In a similar paper, they reported a case of 44 y male medically free who presents with left lower abdominal pain, nausea, fever and peritoneal irritation signs, acute appendicitis was suspected, and no further imaging was required. The decision to go for laparoscopic exploration was made, and the patient was taken to surgery. During their exploration, they found the abnormal position of the stomach at the right and the liver at the left. In the right iliac fossa, the sigmoid colon was occupying the space with multiple inflamed diverticulaes. While in the left iliac fossa, cecal and appendiceal agenesis was found. Tissues were inflamed so lavage was attempted, and they aborted the surgery. Post operative investigations included CT scan. Which confirmed the diagnosis of SIT. In this paper SIT was also associated with different presentation of appendix. Appendicular agenesis is a condition where the development of the vermiform appendix is halted, leading to its complete absence. These cases are typically unexpected during surgery and occur in approximately 0.001% of patients undergoing surgery for suspected acute appendicitis [1].

Regarding the appropriate surgical technique in dealing with patients diagnosed with anatomic anomalies. Laparoscopic exploration is a useful tool when clinical and radiological findings are inconclusive, especially when the appendix is in an unusual anatomical location. This method eliminates the need for large incisions and provides easier access for the trained surgeon [4] Laparoscopy allows for a thorough examination of the entire abdominal cavity, helps to confirm the initial diagnosis and identify any other pathological conditions [6]. There is no fixed protocol for trocar placement in these unusual cases, and the surgeon should adjust port positioning based on key laparoscopic principles, such as triangulation and ergonomics.

4. Conclusion

In conclusion, we suggest detailed history taking and examination in patients with acute abdomen and unclear diagnosis. Always remember to keep a broad differential diagnosis in mind. We recommend screening for any cardiac anomalies in patients with SIT since congenital heart disease is observed in 3% to 5% of these cases [4]. And lastly, to not hesitate with liberal use of laparoscopic exploration as a diagnostic tool in unclear cases of abdominal pain.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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