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International Journal of Surgery Case Reports

journal homepage: www.casereports.com

A colovesical fistula with a persistent descending mesocolon due to partial situs inversus: A case report



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ARTICLE INFO

Article history:

Received 15 March 2017

Received in revised form 13 June 2017

Accepted 13 June 2017

Available online 17 June 2017

Keywords:

Partial situs inversus

Colovesical fistula

Persistent descending mesocolon

ABSTRACT

INTRODUCTION: Situs inversus viscerum, a congenital condition in which the visceral organs are a mirror image of their normal physiological positions, could be total or partial. Persistent descending mesocolon (PDM) is a congenital anomaly that is asymptomatic because of its short length. PDM causing intestinal obstruction is a known clinical complication.

PRESENTATION OF CASE: A 74-year-old woman presented with pneumaturia and enteruria for two months, and recurrent cystitis for a month. An enhanced computed tomography (CT) showed air in the bladder along with sigmoid colonic diverticula adherent to it, suspecting a fistula. The CT also showed partial situs inversus with the common hepatic artery, and left colic artery arising abnormally from the superior mesenteric artery (SMA). Minimally invasive endoscopic closure using the over-the-scope clipping system was difficult because of thickening and scar tissue due to chronic inflammation from diverticulitis. Thus, a sigmoidectomy was performed to close the fistula. Intraoperatively, we noted an abnormally fixed descending mesocolon. An emergency reoperation was performed on the sixth postoperative day owing to an anastomotic leak. Suture failure was attributed to these congenital abnormalities due to insufficient blood flow from an absent marginal vessel and a high endocolonic pressure by adhesions. Sigmoid colon re-resection and maturation of an ileostomy was performed. The patient had no specific postoperative complications, and the ileostomy was closed after three months.

CONCLUSION: We report an extremely rare case of colovesical fistula due to a PDM in a patient having partial situs inversus with abnormal branches originating from the SMA.

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1. Introduction

In patients having situs inversus, the viscera could be present in all locations between their normal and mirror image positions. Situs inversus is a lateralization anomaly often associated with other malformations that alter prognosis such as isolated cardiac malformations, cardiopulmonary syndromes, or Kartagener syndrome [1]. Genetic and environmental factors have been reported as possible causes. Lee et al. report that some cases could be attributed to factors in early embryonic life [2]. Failure of fusion between the primitive dorsal mesocolon and the parietal peritoneum during the

fifth month of gestation leads to PDM. In the 1960s, this abnormality was reported by both Radiology and Gynecology [3,4]. A colovesical fistula is a well-known complication in many different diseases. Among the causes described in literature, the commonest are diverticular and inflammatory bowel disease (predominantly Crohn's) [5,6]. We report an extremely rare case of colovesical fistula due to a persistent descending mesocolon in a patient having partial situs inversus.

2. Presentation of case

A 74-year-old woman known to have renal stones presented to the Department of Urology with a two-month history of pneumaturia and enteruria, and recurrent cystitis for a month. Cystoscopic examination showed an edematous posterior bladder wall with debris flowing from a fistula in the center (Fig. 1a). An enhanced CT scan showed air in the bladder and diverticula adherent to

Abbreviations: PDM, persistent descending mesocolon; SMA, superior mesenteric artery; CHA, common hepatic artery; LCA, left colic artery; CT, computed tomography; OTSC, over-the-scope clipping; IMA, inferior mesenteric artery.

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<http://dx.doi.org/10.1016/j.ijscr.2017.06.029>

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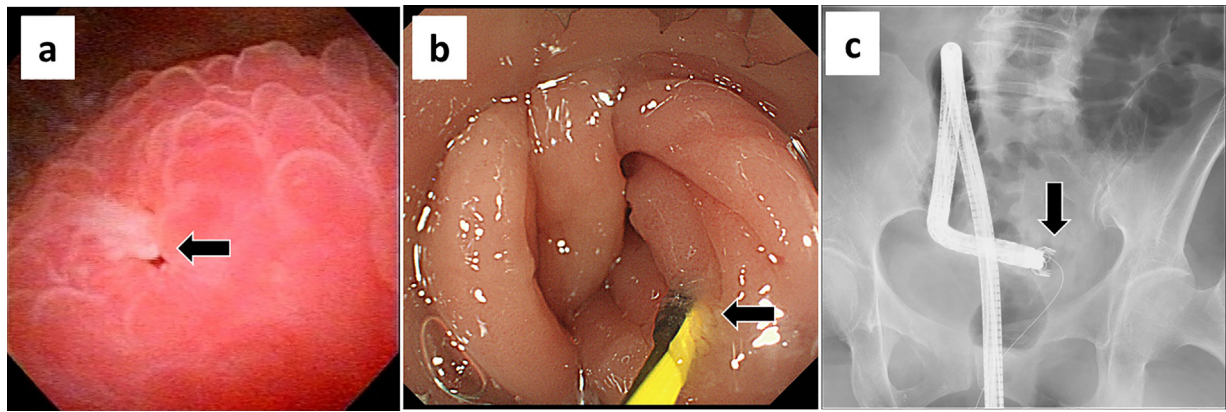


Fig. 1. (a) Cystoscopy showing edematous mucosa of the posterior bladder wall and a hole in the center (arrow). (b) Endoscopy showing the guide wire passing through the colovesical fistula (arrow). (c) Roentgenoscopy showing the endoscope with a mounted OTSC clip (arrow) approaching the fistula.

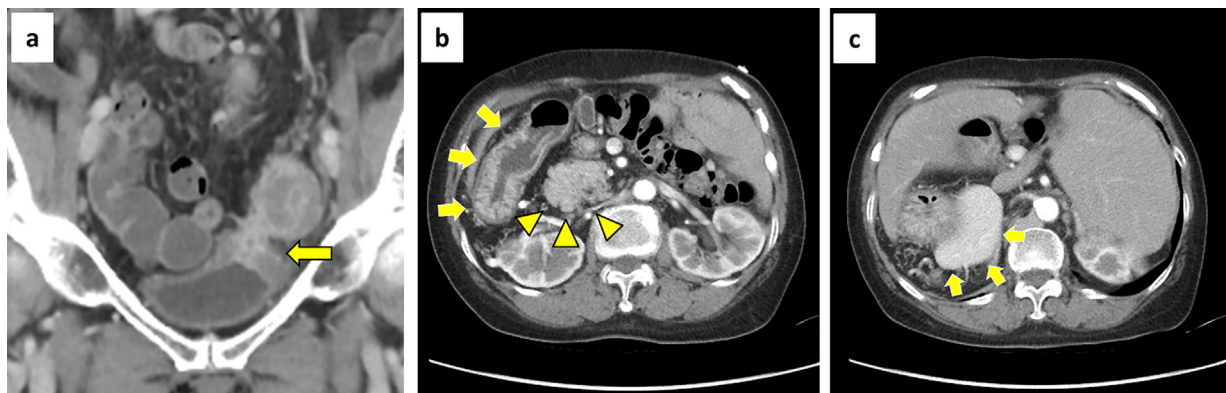


Fig. 2. (a) Coronal CT images showing a thickened wall of the sigmoid colon adherent to the bladder (arrow), and a suspected colovesical fistula. (b) Abdominal CT showing stomach (arrow) and pancreas (arrowhead) transposed to the right side. (c) Abdominal CT showing the spleen on the right (arrow).

it, leading to the suspicion of a fistula (Fig. 2a). Based on these findings, she was diagnosed as having a colovesical fistula due to sigmoid diverticulitis. The CT scan also revealed transposition of

the intra-abdominal organs, viz., the stomach, spleen, and pancreas with the descending colon transferred to the middle of the pelvis (Fig. 2b and c). Additionally, it also showed the common

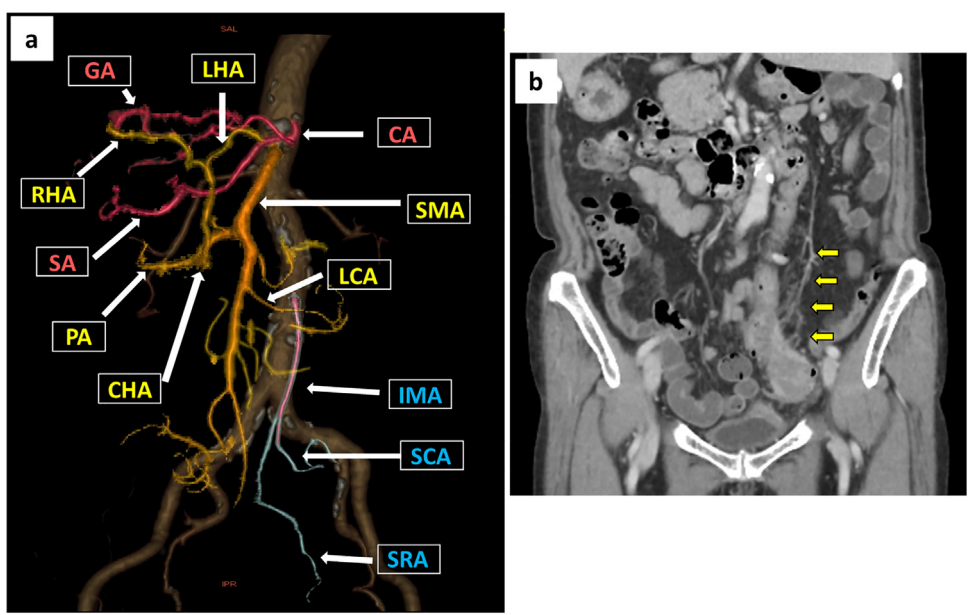


Fig. 3. (a) Common hepatic artery and left colic artery arising from the superior mesenteric artery. (CA: celiac artery, SA: splenic artery, GA: gastric artery, SMA: superior mesenteric artery, CHA: common hepatic artery, RHA: right hepatic artery, LHA: left hepatic artery, PA: pancreatic artery, LCA: left colic artery, IMA: inferior mesenteric artery, SCA: sigmoid colon artery, SRA: superior rectal artery). (b) Sigmoid artery and superior rectal artery branching from the inferior mesenteric artery (arrow).

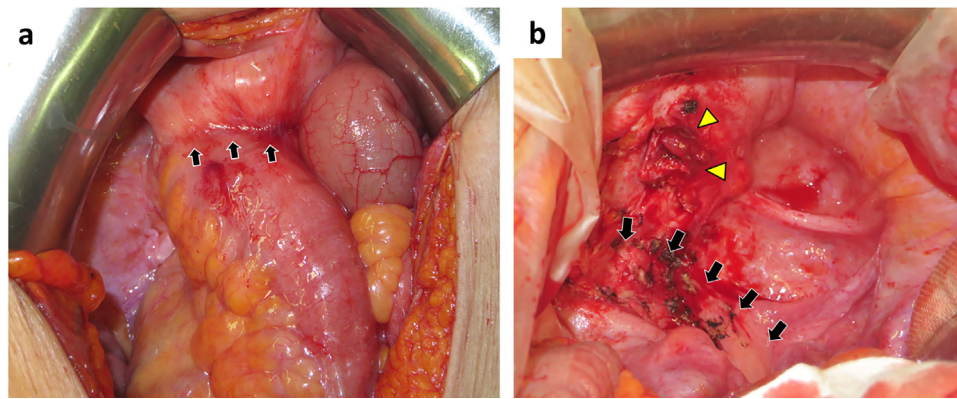


Fig. 4. (a) This is a picture at the time of laparotomy. The sigmoid colon is strongly adherent to the bladder (arrow). (b) Intraoperative findings showing an adhesion and shortening at the dorsal aspect of the descending and sigmoid mesocolon (arrow). Also showing exposure of the bladder fistula (arrowhead).

hepatic artery (CHA), and left colic artery (LCA) arising abnormally from the SMA, and the inferior mesenteric artery (IMA) going to the sigmoid colon and rectum without marginal vessels (Fig. 3a and b). We attempted a minimally invasive endoscopic closure of the fistula using the over-the-scope clipping (OTSC) system. A guide wire was inserted through a hole in the bladder into the sigmoid colon (Fig. 1b). The endoscope with an applicator cap attached to its tip and the mounted OTSC clip, was placed into the fistula using the guide wire (Fig. 1c). However, it was difficult to close the fistula using the OTSC system because of thickening and scar tissue due to chronic inflammation from diverticulitis. Therefore, a surgical resection was performed. The descending and sigmoid colon were noted to be fixed to the retroperitoneum by an adhesion of its mesocolon (Fig. 4a and b). After lysing this adhesion, a sigmoid colectomy and bladder repair were performed, and the colon was reconstructed using hand-sewn end-to-end anastomosis. Drainage catheters were placed in the pelvis. Histological examination revealed a colovesical fistula with acute inflammation and fibrosis (Fig. 5a and b).

On the fourth postoperative day, she spiked a high fever and developed abdominal pain. Because of the turbid fluid in the drain, we diagnosed her as having an anastomotic leak, but elected conservative management because her general condition was stable and a CT scan showed an area of limited leakage. However, on the sixth postoperative day, her abdominal pain worsened and some liquid feces were noted in the drainage tube. She therefore underwent emergency surgery. The posterior side of the anastomosis was completely separated and ischemic changed. Additionally, sigmoid colon resection and maturation of a double barrel ileostomy were carried out in the right lower quadrant of the abdomen. The patient had no specific postoperative complications. She was discharged 30 days postoperatively, and the ileostomy was closed after three months.

3. Discussion

Total or partial situs inversus viscerum, is a congenital condition where the position of organs is a mirror image of their normal physiological positions [1]. The incidence of situs abnormalities is unknown. Some studies estimate that it occurs in approximately 1 per 8000–25,000 live births [7]. Partial situs inversus is rarer than situs inversus totalis [8]. The pathogenesis of situs inversus is not yet clear. In the partial type, there may be only thoracic inversion, cardiac chamber reversal, or only abdominal organ inversion accompanied by syndromes showing splenic anomalies, an annular pancreas, a horseshoe kidney, a diaphragmatic hernia, or other developmental abnormalities [7]. The partial situs inversus described was probably caused by a reversal of location in terms of right and left, of the duodenum, pancreas, liver, stomach, and spleen, due to a possible abnormality in rotation in early embryonic life [9].

In the present case, a CT revealed inversus of the intra-abdominal organs, viz., the stomach, spleen, and pancreas with the descending colon transferred to the middle of the pelvis. There was also an abnormal origin of the CHA and LCA from the SMA with the IMA going to the sigmoid colon and rectum without marginal vessels. Although malrotation of the intestines is often associated with situs inversus, it was not so in our case [10,11]. CHA originating from the SMA is rare and accounts for only 1.5% of all cases [12]. It corresponds to a Type V variant per Hiatt's classification. Hiatt JR et al. have classified the hepatic artery anatomy into 6 types (Type I: normal configuration, Type II: replaced or accessory left hepatic artery arising from the left gastric artery, Type III: replaced or accessory right hepatic artery arising from the SMA, Type IV: replaced or accessory right hepatic artery arising from the SMA, Type V: CHA arising from the SMA, Type VI: CHA arising from the aorta). The left colic artery arises from the SMA in only 1% of all

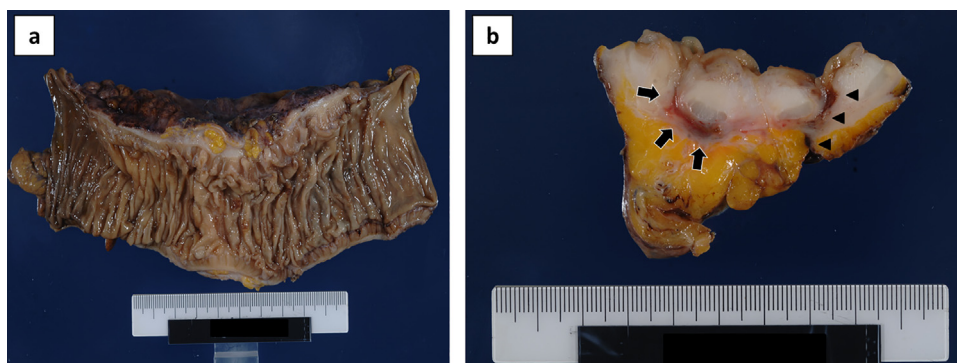


Fig. 5. (a) The picture shows a resected specimen. (b) Picture of the cut surface showing acute diverticulitis and fistula.

cases [13]. The incidence of these two anomalies in an individual is extremely low (approximately 1.5×10^{-4}), if they occur independently.

A colovesical fistula is a well-known complication in several different diseases. Rufus of Ephesus first reported it in AD 200 [14]. As mentioned in a previous study, diverticular disease is a major cause of benign colovesical fistulae [6]. Open surgical procedures remain the mainstay of treatment for colovesical fistulae [15]. Initially, we tried a minimally invasive endoscopic closure of the fistula using the OTSC system, which has made possible the closure of gastrointestinal tract defects, such as perforations, anastomotic insufficiency, and fistulae, as well as achievement of hemostasis in difficult-to-treat non-variceal bleeding [16,17].

In our case, the OTSC system failed because of thickening and scar tissue due to chronic inflammation from diverticulitis, so we performed a sigmoid colostomy to close the fistula. The hand-sewn suture anastomosis unfortunately ruptured on the fourth postoperative day, and we attribute the failure of the sutures to PDM, a congenital anomaly that, in most cases, is asymptomatic because of its short length [18]. In our case, a CT scan showed the descending colon shifted to the midline of the abdomen with radial branches from the inferior mesenteric artery instead of a marginal vessel. Moreover, intraoperatively, there were adhesions noted at the dorsal aspect of the descending and sigmoid mesocolon—typical of PDM [19]. PDM causing intestinal obstruction is a known clinical complication [20]. Considering all the above-mentioned facts, we inferred that suture failure occurred from an insufficient blood flow following absence of marginal vessels, and high endocolonic pressure due to these congenital abnormalities such as partial situs inversus viscerum and PDM.

4. Conclusion

We report an extremely rare case of a colovesical fistula with a PDM in a patient having partial situs inversus with abnormal branches originating from the SMA. In such cases, some attention needs to be directed toward maintaining intestinal blood flow and the management of adhesions to avoid failure of sutures.

Conflicts of interest

None of the authors has anything to disclose.

Sources of funding

None of the authors has anything to disclose.

Ethical approval

All procedures used in this research were approved by the Ethical Committee of Chugoku Rosai Hospital.

Consent

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Author contribution

Fukuhara, Tazawa, Takahashi and Sakimoto wrote the manuscript. Kuga and Miwata diagnosed this case. Tazawa, Hirata, Fukuhara, Imaoka, Fujisaki and Fukuda performed the operation. Nishida diagnosed this disease pathologically. All authors conceived of the study and participated in its design and coordination and helped to draft the manuscript. All authors read and approved the final manuscript.

Guarantor

Hirofumi Tazawa has accepted full responsibility for this work and the decision to publish it.

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