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Laparoscopic Cholecystectomy with a Mixed Approach in a Patient with Kartagener Syndrome: Technical Report and Review of Literature

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Abstract

Kartagener syndrome (KS) is a rare autosomal recessive disease. The disease is characterized by three typical symptoms: chronic sinusitis, situs viscerum inversus (SVI), and bronchiectasis. The laparoscopic cholecystectomy (LC) is the standard procedure in most cases of cholelithiasis, but in SVI patients it can be difficult, especially for right-handed surgeons. We report the case of a 24-year-old female affected by KS, presenting with a history of symptomatic cholelithiasis. Ultrasound and magnetic resonance cholangiopancreatography confirmed SVI totalis and cholelithiasis. The patient underwent a laparoscopic cholecystectomy by a right-handed surgeon performed with a mixed approach without complications. Laparoscopic cholecystectomy in SVI patients can be a safe and reliable technique especially for a left-handed surgeon. The described technique is also easy for a right-handed surgeon. However, it is considered a technically challenging procedure and often requires technical modification.

Keywords Situs viscerum inversus · Laparoscopic cholecystectomy · Kartagener syndrome · American mirror technique · French mirror technique

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Introduction

Kartagener syndrome (KS) is a rare autosomal recessive disease, described for the first time by Siewert in 1904, even if Kartagener clinically recognized the syndrome in 1933 for the first time [1]. This condition represents a subgroup of primary ciliary dyskinesia (PCD), a group of rare inborn pathologies clinically and genetically heterogeneous caused by a mutation occurring on many different genes modifying the structure and/or the function of motile cilia in different tissues. PCD prevalence is 1:15000/1:30000 and the KS represents 50% of them [2–4]. The disease is characterized by three typical symptoms: paranasal sinuses chronic infections, situs viscerum inversus (SVI), and bronchiectasis [5]. There is no evidence of a relationship between gallbladder stones and KS; it is assumed that cholelithiasis occurs with the same frequency of general population [6]. The laparoscopic cholecystectomy (LC), introduced by Mouret in 1987, represents the standard procedure in most of cholelithiasis cases. However, in SVI patients, LC can be difficult, especially for right-handed

surgeons [7]. The first case of LC in SVI patient reported in literature dates to 1991 [8] and up to now there are about 136 reported cases [9]. We present a case of LC in a patient with KS and SVI totalis and performed a review of the literature about this topic.

Case Report

A 24-year-old female, affected by KS detected with the classical triad of symptoms (bronchiectasis, chronic sinus infections, and SVI totalis) presented with a history of symptomatic cholelithiasis. The patient had never undergone surgery and she referred the absence of any comorbidities. She complained nausea associated with epigastric and left hypochondrium pain with posterior irradiation worsening after meals in the last three months. She had no fever, no weight loss, or jaundice. At physical examination, we found mild epigastric discomfort on palpation. Liver function tests and complete blood count were normal. Chest X-ray (Fig. 1) showed dextrocardia and fundus of stomach positioned in the right abdominal region. Ultrasound evaluation confirmed SVI totalis with liver and gallbladder (with multiple stones) in the left upper quadrant and spleen in the right upper quadrant. Magnetic resonance image (MRI) and magnetic resonance cholangiopancreatography (MRCP) showed no intra- and extrahepatic biliary ducts dilations and no anatomical abnormalities (Fig. 2). Because of the evidence of symptomatic cholelithiasis, the patient underwent elective LC under general anesthesia.

Usually LC is performed with the “French technique”: the patient is positioned supine on the operating table with legs spread apart and surgeon between them; the monitor is placed on the right and the first assistant on the left side of the patient. Differently, the technique mainly adopted in SVI patients is the “American mirror technique,” especially for right-handed surgeons: the surgeon and the first assistant are on the right side, while the monitor and the second assistant are on the left.



Fig. 1 Chest X-ray showing dextrocardia and presence of the fundus of stomach in the right abdominal region

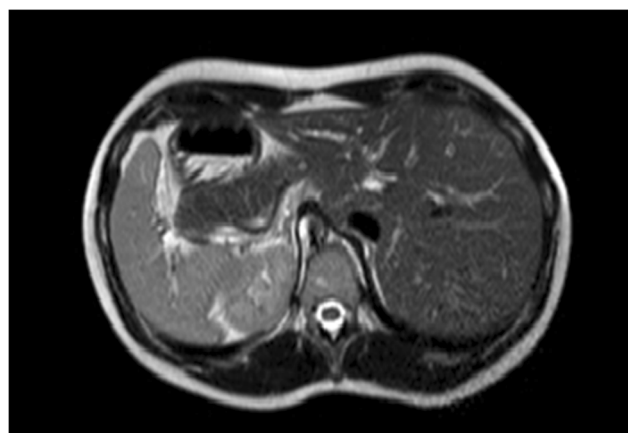


Fig. 2 MRCP showing SVI totalis

To perform the surgery without difficulty with the right hand, we used a combination of the two techniques: patient positioned according to the “French technique,” operator on the right according to the “American” one, first assistant between the spread apart legs, and the monitor on the left. The four trocars were introduced in complete mirror image to the standard “French technique” (Fig. 3). We established pneumoperitoneum by an open technique using a 12-mm blunt optical trocar, through the infraumbilical incision, insufflating carbon dioxide with a pressure of 12 mmHg. The diagnosis of SVI was confirmed at the exploration of the abdominal cavity. We inserted the second 10-mm trocar on left mid-clavicular line below the costal margin, the third 5-mm trocar into the subxiphoid area on the left of the median line, and the fourth 5-mm trocar along the anterior axillary line at the level of the umbilical line (Fig. 3). The patient was positioned in reversed Trendelenburg position and slightly turned to the right. During the first step of surgical procedure, the surgeon used the left mid-clavicular port (second trocar) for dissection with his right hand and the subxiphoid port (third trocar) to retract the Hartmann’s pouch with his left hand. These movements allowed a complete vision and dissection of the posterior peritoneal sheet. In the second step, the first assistant held the 30° camera through the first trocar, and pulled up the Hartmann’s

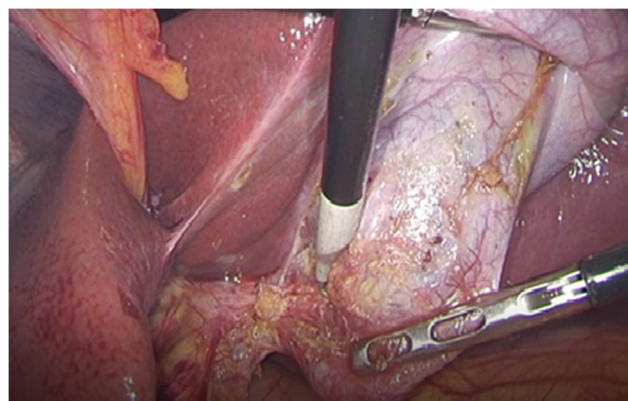


Fig. 3 Positioning of the four trocars

pouch to show the anterior peritoneal sheet through the fourth trocar, while the surgeon released Hartmann's pouch and retracted the fundus of the gallbladder through to the third trocar to continue the dissection through the second one (Fig. 4). Once Calot's triangle was cleared by fat tissue achieving a clear and safe vision, the cystic duct and artery were localized, clipped, and divided. The gallbladder was dissected from the liver plate, placed in an endobag, and removed through the umbilical port. The operation time was 45 min. The postoperative period was uneventful, and the patient was discharged on postoperative day 1. Stitches were removed after 7 days during an outpatient visit. Histopathologic analysis showed a chronic cholecystitis with adenomyosis outbreaks.

Discussion

The KS is an autosomal recessive disorder characterized by bronchiectasis, chronic sinusitis, and SVI totalis. The first description of SVI in animals was made by Aristoteles [10], while the first human case was reported by Fabricius in 1600 [11]. There is no current evidence that SVI is more associated with cholelithiasis and the non-specific symptoms and signs may determine a delay in the diagnosis. Most of the patients with SVI and cholelithiasis present left-sided upper abdominal pain, about 30% of them epigastric pain, and nearly 10% pain in the right hypochondrium [12–14]; probably in these last cases, there is an incomplete inversus of the autonomic nervous system. Regardless of the different localization of pain and clinical presentation of cholelithiasis, the complications are similar [12]. Ultrasonography, abdominal CT, chest RX, and MRI not only confirm SVI totalis but also show the presence of any anatomical anomalies of the biliary tracts.

We searched on PubMed using the following keywords: "Situs viscerum inversus," "Laparoscopic cholecystectomy," "Kartagener Syndrome," "American mirror technique," and "French mirror technique". In this way, we found 40 cases of open cholecystectomies and 136 cases of LC in patients

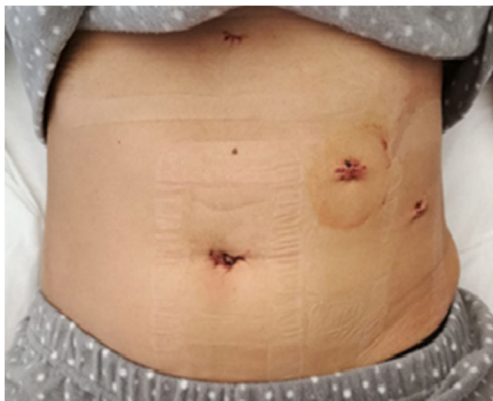


Fig. 4 Position of trocars during gallbladder dissection

with SVI. The first LC in patient with SVI was reported in 1991 by Campos and Sipes [8]. Details about the LC cases are reported in Table 1. In particular, 97 patients had chronic cholecystitis (of them 2 with empyema, 10 with common bile duct (CBD) calculi, 4 with appendicitis, 3 with biliary pancreatitis, 1 with chronic myeloid leukemia, 1 with gastric cancer, 1 with rectal cancer, and 1 undergoing bariatric surgery) and 39 patients had acute cholecystitis (of them 1 with empyema, 3 with CBD calculi) [9, 13, 15]. Like in general population, also in this case series, there is a prevalence of female gender (97 female and 39 males). Then, 129 patients had SVI totalis while 7 SVI partialis. Endoscopic retrograde cholangiopancreatography (ERCP) was used in 12/13 cases of CBD. Data about increase in mortality or higher complication rate (like bile duct injuries and bleeding) are not reported.

All these studies demonstrated reliability and safety of LC [9, 13, 15], even if with some difficulties: (1) it requires a different position for the operating instruments; (2) it is necessary to identify and adapt to the mirror anatomical image. These aspects make the procedure longer than the standard LC [11]. The mirror arrangement of the organs, with respect to their usual position, is not associated with arterial, venous, and extrahepatic bile ducts anomalies in KS [12, 13]. The most commonly adopted surgical technique for LC in SVI patients was the "American mirror," with the monitor positioned on the left side, the surgeon and first assistant on the right side, and the second assistant on the left side of the patient. The position of four trocars is mirrored if compared with the standard LC [7, 9, 13, 16]. This type of procedure was easier for a left-handed surgeon because of the dissection and clips application with the left hand through the xiphoid access, while the first assistant retracted the Hartmann's pouch. In this way, their hands did not cross. However, greater coordination between the two surgeons would be necessary.

To overcome this difficulty, unlike the other authors, we adopted a mixed approach between the French and the American technique: the surgeon placed to the right of the patient and using the right hand through the access to the left mid-clavicular line (second trocar), to dissect; in the first time uses left hand to retract the Hartmann's pouch and in second time the fundus of the gallbladder one, through the subxiphoid port (third trocar). The first assistant is placed between the legs of the patient, maintaining the laparoscope through the first trocar and carrying out, if necessary, the traction of the anterior peritoneal sheet through the fourth trocar. With this technical modification, we obtained a greater easiness of movement, reducing interferences between surgeons with a greater vision of the operative field. In addition, the most important aspect is that the surgeon always uses for dissection and clips application the right hand. However, our mixed approach needs a good coordination between surgeons. We think it should be performed by an experienced surgeon, because mirror arrangement of the organs, compared with their usual

Table 1 List of cases of LC in patients with SVI

	Author	Year of publication	Sex	Age	Diagnosis	SVI
1	Campos and Sipes et al.	1991	F	39	CC	T
2	Takei et al.	1992	F	51	AC	T
3	Lipschutz et al.	1992	M	80	AC/CBD calculi	T
4	Goh et al.	1992	M	62	CC/empyema	T
5	Drover et al.	1992	F	29	CC	P
6	Huang et al.	1992	M	36	CC	T
7	Schiffino et al.	1993	F	53	CC	T
8	Mc Dermott JP et al.	1994	M	66	CC/CBD calculi	T
9	Malatani TS et al.	1996	F	25	AC	T
10	Crosher et al.	1996	M	63	CC	T
11	D'Agata A et al.	1997	F	72	CC	T
12	Habib et al.	1998	F	45	CC/CBD calculi/CML	T
13	Demetriades et al.	1999	F	61	AC	T
14			M	37	CC	T
15	Djohan et al.	2000	F	20	CC/appendicectomy	T
16	Kombarozos V et al.	2000	M	61	AC/empyema	T
17	Wong J et al.	2001	F	68	CC/CBD calculi	T
18	Donthi et al.	2001	F	43	CC	P
19			F	34	CC	T
20	Nursal et al.	2001	F	42	CC	T
21	Yaghan et al.	2001	F	48	CC	T
22			F	38	CC	T
23	Al Jumaily M et al.	2001	F	46	CC	T
24	Franklin et al.	2001	F	25	CC/appendicectomy	T
25	Singh and Dhi et al.	2002	F	42	CC	T
26	Trongue A et al.	2002	F	28	CC	P
27	Polychronidis et al.	2002	M	68	CC	T
28	Oms and Badia et al.	2003	F	70	AC	T
29			M	65	AC	T
30	Jesudason et al.	2004	M	69	CC	T
31	Kang and Han et al.	2004	F	64	CC/CBD calculi	T
32	Docimo et al.	2004	F	41	CC	T
33	Pitiakoudis et al.	2005	F	47	CC	T
34	McKay D et al.	2005	F	32	AC	T
35	Kamitani et al.	2005	M	76	AC	T
36	Puglisi et al.	2006	F	43	CC	T
37	Bedioui et al.	2006	F	58	AC	T
38	Aydin et al.	2006	M	35	CC/CBD calculi	T
39	Kirshtein B et al.	2006	F	51	AC	T
40	Norman Machado et al.	2006	F	65	CC	T
41	Shah AY et al.	2006	F	60	CC	T
42	Kumar et al.	2007	F	57	CC	T
43	Fernandes et al.	2008	F	43	CC	T
44	Hamdi J et al.	2008	M	41	AC	T
45	Pavlidis TE et al.	2008	F	34	AC	T
46	García-Núñez et al.	2008	F	37	CC	T
47			F	39	CC	T
48	Rosen et al.	2008	M	36	AC	T
49	Paresta I et al.	2008	F	35	CC	T
50	Taskin et al.	2009	F	20	CC/bariatric surgery	T
51	Masood et al.	2009	F	42	CC	T
52	Pereira-Graterol et al.	2009	F	70	CC	T
53	Romano et al.	2009	F	67	CC	T
54	Eisenberg et al.	2009	M	61	AC	T
55	Ghosh N et al.	2009	F	27	AC	T
56	Velimezis G et al.	2009	F	66	CC	T
57	Simmons JD et al.	2009	F	18	CC	P
58	Pataki et al.	2010	F	68	CC	T
59	Hall et al.	2010	M	53	CC	T
60	Gonzalez Valverde et al.	2010	F	46	CC	T

Table 1 (continued)

	Author	Year of publication	Sex	Age	Diagnosis	SVI
61	Sandu C et al.	2010	F	64	CC	T
62	Patle NM et al.	2010	F	36	CC	T
63			F F	43	CC CC	T T
64			F	27	CC	T
65			F M	48	CC AC	T T
66				59		
67				33		
68	Jindal et al.	2010	F	55	AC	T
69			F	26	AC	T
70	Sardar H Arif et al.	2010	F	42	CC	T
71	Han et al.	2011	M	45	CC	T
72	Ozsoy et al.	2011	F	65	AC	T
73	Uludag et al.	2011	M	49	AC	T
74	Borgaonkar et al.	2011	F	47	CC/appendicectomy	T
75	Weber-Sánchez A et al.	2011	M	60	AC	T
76	Seo KW et al.	2011	M	60	CC/gastric cancer	T
77	Evoli LP et al.	2011	F	48	CC	T
78	Iusco et al.	2012	F	52	CC	T
79	Elbeshry et al.	2012	F	24	CC	T
80	Lochman et al.	2012	F	75	AC	T
81	Demiryilmaz I et al.	2012	F	55	CC/CBD calculi*	T
82			M	51	CC	T
83	de Campos Martins, Marcus Vinicius Dantas et al.	2012	F	59	CC	T
84	Pahwa et al.	2012	F	46	CC	T
85	Bozkurt et al.	2012	M	49	CC	T
86	Pezzolla et al.	2012	F	36	AC/pregnancy	T
87	Elbeshry et al.	2012	F	24	CC	T
88	Sandhu S et al.	2012	M	17	AC	P
89	Evoli LP et al.	2012	F	48	CC	T
90	Salama IA et al.	2013	M	10	CC	T
91	Singh V et al.	2013	F	45	AC	T
92	Arya et al.	2013	F	35	CC	T
93	Ali MS et al.	2013	F	43	CC	T
94	Khiangte et al.	2013	M	65	CC	T
95	Stojcev et al.	2013	M	47	CC	T
96	Zhang SN et al.	2013	F	50	AC	T
97	Raghuvveer et al.	2014	M	55	CC	T
98	Reddy A et al.	2014	F	45	AC/CBD calculi	T
99	Mn R et al.	2014	F	55	CC	T
100	Fang et al.	2015	F	39	CC/rectal cancer	T
101	Deguchi Y et al.	2015	M	66	CC	T
102	Phothong et al.	2015	F	39	CC	T
103	Butt MQ et al.	2015	F	40	CC	T
104	EL Saady AM et al.	2015	F	52	CC	T
105	Alsebek MB et al.	2016	F	50	CC	T
106	Ahmed et al.	2016	F	46	AC	P
107			F F	44	CC CC	T T
108				33		
109	Goyal et al.	2016	F	52	CC	T
110			F	50	CC	T
111	Bothra R et al.	2016	F	45	CC	T
112	Berbers F et al.	2016	M	44	AC	T
113	Duncan TK et al.	2016	F	43	AC/CBD calculi	T
114	Jian-jun Ren et al.	2017	F	36	CC	T
115	Kafadar et al.	2017	F	59	AC	T
116	Haddad A et al.	2017	M	58	AC	T
117			M	54	AC	T
118	Centeno DM et al.	2017	F	16	CC	T
119	Lutsevich O et al.	2017	M	75	CC	T
120	Ahmad R et al.	2017	F	57	CC/biliary pancreatitis	T

Table 1 (continued)

	Author	Year of publication	Sex	Age	Diagnosis	SVI
121	Rungsakulkij et al.	2017	M	32	CC/biliary pancreatitis	T
122	Fanshawe AEE et al.	2017	F	53	CC/biliary pancreatitis	T
123	Alam et al.	2017	F	20	CC	T
124	El Hajj et al.	2017	M	61	CC/biliary pancreatitis/CBD calculi	T
125	Ying et al.	2017	F	51	AC	T
126	Ren J et al.	2017	F	36	CC	T
127	Yogesh et al.	2018	F	50	CC/CBD calculi	T
128	Jhobta RS et al.	2018	F	23	CC	T
129	Takalkar et al.	2018	F	50	CC/CBD calculi	T
130	Žatecký et al	2018	F		CC/empyema	T
131	Hu L et al.	2018	M	72	CC/CBD calculi	T
132	Boufettal et al.	2018	M	25	AC	T
133	Petit et al.	2018	F	33	AC	P
134	Omar Alkhlaify et al.	2019	M	40	CC	T
135	Mohammed A et al.	2019	M	26	AC	T
136	Malik FS et al.	2019	F	40	AC	T
137	Current study	2019	F	24	CC	T

AC, acute cholecystitis; CC, chronic cholecystitis; M, male; F, female; P, partialis; T, totalis; CML, chronic myeloid leukemia; CBD, common bile duct

*The authors did not report ERCP in this case of CBD calculi

position not only influences the localization of symptoms and signs during the clinical approach but also imposes preoperative careful and surgical technique planning. Romano et al. in 2009 described a similar technique but with one difference: to create a clear operative field, they inserted a fifth 5-mm trocar on the anterior axillary line of the left hypochondrium [17]. Furthermore, the operative time was 120 min due to the anatomical peculiarity and the difficulty of this not standardized surgical procedure [17]. Patle et al. and Ghosh et al. proposed the LC with the positioning of the surgeon between the patient's legs, first assistant on the right, and second assistant and monitor on the left [16, 18]. They positioned the trocars in a mirror image compared with the standard technique. In our opinion although the surgeon operates on with his right hand, this position does not allow him to be able to move easily with his left hand. Additionally, there are some difficulties because of the instruments crossing increasing operative time (from 45 to 85 min) [16, 18]. The increased operative time and the pneumoperitoneum can lead to intra- and postoperative complications due to the comorbidities of patients with Kartagener syndrome. In order to allow the surgeon to dissect with his right hand, Ali et al. proposed to stay at the patient's right, positioning one of the 5-mm trocars on the right mid-clavicular line instead of the xiphoid line and the 10-mm trocar on the left mid-clavicular line [19]. Even in this case, the method was safe but with an increase of the operative time [19]. In a systematic review, Chaouch et al. concluded that LC is considered an undoubtful treatment for cholelithiasis even in SVI patients, easier for left-handed surgeons but with the "American mirror technique" fastest also for right-handed surgeons. Some modifications of the port placement can facilitate

this last procedure [9]. Most of the studies describe LC in patients with SVI with four standard accesses, but there are papers describing LC with three trocars [20] and with a single access [21–23]. Iusco et al. placed three trocars using the "French technique": a 10-mm sub-umbilical trocar, a second 10-mm in the upper right quadrant, and one of 5-mm in the right flank [20]. The limitation of this procedure is that the surgeon dissected with the left hand, a difficult maneuver for a right-handed operator causing longer operative time [20]. In 2011, Han et al. reported a single access procedure in a patient with SVI [21]. The authors described the procedure as safe and reliable and associated with less postoperative pain; moreover, intersecting the instruments, the difficulties encountered by a right-handed surgeon are overcome since he dissects with his right hand [21]. In 2015, Deguchi et al. defined LC with a single access in SVI patient as a more difficult procedure than traditional LC, due to associated technical problems, including the crowding of laparoscope and other instruments in the umbilical port [23]. This situation caused interferences between instruments inside the abdomen and even outside, the loss of their triangulation in the operative field, pneumoperitoneal leakage, intra-abdominal smoke, and longer operating times to allow a precise and safe dissection [23].

Conclusions

Laparoscopic cholecystectomy in patients with cholelithiasis and situs viscerum inversus totalis could be considered as a safe and reliable technique, especially for a left-handed surgeon. Our technical procedure is also easy for a right-handed

surgeon. However, we think it should be performed by an experienced surgeon, because the different anatomy of the gallbladder not only influences the localization of symptoms and signs but also imposes a correct preoperative planning with the appropriate technical devices and a greater surgeon's ability.

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