CASE REPORT

Laparoscopic sleeve gastrectomy in a patient with *situs inversus totalis* and Kartagener syndrome: an unusual surgical conundrum

Angela Burvill,¹ Ruth Blackham,^{2,3} Jeffrey Hamdorf^{2,3}

¹Medical School, University of Western Australia, Crawley, Western Australia, Australia

²School of Surgery, University of Western Australia, Crawley, Western Australia, Australia

³Hollywood Private Hospital, Western Surgical Health, Nedlands, Western Australia, Australia

Correspondence to

Miss Angela Burvill,
21486025@student.uwa.
edu.au

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SUMMARY

Kartagener syndrome is a rare autosomal recessive condition. Approximately 25% of those with *situs inversus totalis* suffer the syndrome. With the rising overall number and indications for bariatric surgery, this condition will be increasingly recognised. We present a case of a 25-year-old woman with *SIT* and Kartagener syndrome who underwent a laparoscopic sleeve gastrectomy. As with all bariatric surgery, a multidisciplinary team approach was important in managing such a case. There were considerable cognitive challenges for the surgical team both preoperatively and during the procedure. The patient tolerated the operation well and was discharged 2 days after the surgery. At 12-months follow-up, the patient had achieved 125% excess weight loss. This case illustrates that an experienced surgeon can safely perform a laparoscopic sleeve gastrectomy on a patient with *situs inversus totalis*.

BACKGROUND

Bariatric surgery is the most effective treatment for clinically severe obesity, with substantial and sustained effects on weight reduction and improvement in obesity-related comorbidities.^{1–3} Laparoscopic sleeve gastrectomy (LSG) has been increasingly accepted as a primary bariatric intervention,⁴ and its effectiveness in percentage excess weight loss exceeds laparoscopic adjustable gastric banding.

Laparoscopic surgery in patients with *situs inversus totalis* (*SIT*) can be technically demanding for gastrointestinal surgeons.⁵ *SIT* is a rare congenital condition where the intrathoracic and intra-abdominal organs are completely reversed along a central axis. The incidence of *SIT* is estimated at 1:5000–20,000 newborns.⁶ The reversal of the placement of intra-abdominal structures requires cognitive and manual dexterity on the part of the surgeon to predict and manoeuvre accordingly.

Approximately 25% of those with *SIT* have Kartagener syndrome.⁷ The syndrome is defined as the triad of bronchiectasis, sinusitis and *SIT*. Kartagener syndrome occurs in approximately 50% of patients with primary ciliary dyskinesia.⁷ Primary ciliary dyskinesia is an autosomal recessive disease characterised by ciliary dysfunction and impaired mucociliary clearance, resulting in an array of pulmonary and other manifestations.

CASE PRESENTATION

A 25-year-old woman with Kartagener syndrome had a preoperative weight of 116 kg (body mass index of 40 kg/m²). By the definition of Kartagener syndrome, the patient had *SIT*, which included within the syndrome the conditions of bronchiectasis and sinusitis. The patient had been warned, and due to her inherent defective cilia, any future pregnancy may be more difficult as a result. She was a non-smoker with occasional alcohol consumption only. She was lactose intolerant but without other comorbidities.

The patient reported being overweight since the age of 16 years and had tried unsuccessfully for many years to lose weight. She elected to have a LSG for morbid obesity.

The patient's sinusitis and bronchiectasis made her prone to upper respiratory tract infections, for which she required regular courses of antibiotics. The patient's last course of antibiotics was 2 days prior to the surgery, which placed her at a greater risk of postoperative pneumonia.

At the surgical centre, a multidisciplinary approach was taken. In the lead-up to the operation, the patient was required to consult the bariatric surgeon, along with a bariatric general practitioner, a psychologist and a dietician. The patient in this case had bronchiectasis and so was required to consult a respiratory physician. The physician then advised the medical and surgical team about reducing the patient's risk of pulmonary complications during and after the operation.

TREATMENT

The patient was scheduled for an elective LSG. The patient's bronchiectasis and *SIT* posed unique challenges to the medical and surgical team.

The anaesthetic team adopted its usual protocol to the bariatric surgery and did not alter its approach to the induction or maintenance of the anaesthesia.

The procedure included all of the standard surgical steps for LSG, performed in the mirror position. The surgeon and scrub nurse were positioned on opposing sides to their usual positioning such that the surgeon stood on the patient's right side. The port placement is very important in such cases, as a working port too lateral or high can make an already challenging case even more so. The port placements were reversed as shown (figure 1). The spleen was visualised in the right upper quadrant,

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Figure 1 Port set-up.

and the gastrocolic ligament and short gastric vessels were then ligated and divided. A stay suture applied to the greater omentum adjacent to the gastric fundus permitted downward traction, enhancing the view of the proximal stomach, left crus and gastro-oesophageal region. The greater omentum was divided from the patient's left towards the right, and the stapling device applied in the reversed left-to-right configuration (*figure 2*). The gastric pouch was removed through the right lateral working port. Five cartridge loads (two black and three purple) of the stapler were used (EndoGIA Reinforced Reload with Tristaple technology; Medtronic), and a 38 Fr bougie was used to ensure a satisfactory gastric remnant size. The first staple cartridge was applied some 4 cm from the pylorus, and the proximal extent

Figure 2 Incision from right to left, the linear stapler held in the opposite direction from the usual.

of the staple line was within 1 cm of the gastro-oesophageal junction.

In terms of non-technical challenges, surgeons must be aware of the difficulties that may arise in overcoming cognitive dissonance associated with *SIT* patients. Cognitive dissonance concerns situations in which individuals have two cognitive pathways that are relevant to each other but seemingly inconsistent.⁸ The mirrored arrangement of the intra-abdominal organs may be disconcerting to gastrointestinal surgeons.

OUTCOME AND FOLLOW-UP

The procedure was completed successfully. The operative time was 35 min, slightly longer than usual. The patient recovered uneventfully and was discharged 2 days later. After two weeks, the patient had decreased from operative weight of 116–104 kg and was tolerating nourishing fluids before transitioning onto a puree diet. At review, 6 weeks following surgery, she had progressed to a normal texture diet. Her weight at that review was 96 kg (17% total weight loss). At her most recent review, 12 months from the date of surgery, she had reached a weight of 64 kg (45% total weight loss).

DISCUSSION

In our review of the literature, we identified 27 cases of laparoscopic bariatric surgery on *SIT* patients. The published English language medical literature on laparoscopic gastric banding (LGB), laparoscopic Roux-en-Y gastric bypass (LRYGB) and LSG, combined with *SIT*, was accessed using MEDLINE, Google Scholar and the institution-specific search utility, OneSearch. A total of 25 articles with 27 cases were identified. These comprise 6 cases that underwent LGB,^{5–9,13} 9 LRYGB^{5,14–21} and 12 LSG.^{22–32} Among these cases, only one case of Kartagener syndrome was identified for each LGB,¹¹ LRYGB¹⁶ and LSG.²⁶ Genser *et al*²⁶ present a video report of a transumbilical single-incision LSG in a patient with Kartagener syndrome. However, multiple-incision surgery for sleeve gastrectomy is evidently more commonly performed.

The literature suggests that experienced laparoscopic bariatric surgeons can perform LGB, LRYGB and LSG safely.^{5,11,13,22–25,27,32} In all cases but two,^{12,24} the operations were performed successfully and without complications. In Deutsch *et al*²⁴'s report, the patient experienced a sleeve leak. The authors believe that this occurred independently of the altered anatomy. In Samaan *et al*¹²'s report on LGB, the patient developed a band erosion at 4 months. The authors comment that this was more likely due to the patient's more posteriorly placed and less mobile fundus than his *SIT*. In the limited number of cases available in the literature, the addition of *SIT* thus does not appear to increase the risk of complications in laparoscopic bariatric surgery.³² There were no complications experienced by the patient in this case.

The literature identifies the importance of the preoperative diagnosis of *SIT*, the establishment of associated anomalies and preoperative planning for the technical and nontechnical challenges of surgery to help reduce the risk of complications in *SIT* patients.

The patient may not be aware of the *SIT*, and therefore, the medical and surgical teams should be alert to the possibility of condition in the preoperative work-up. Samaan *et al*¹² comment that there would be no justification to change the standard work-up to detect *SIT*, given its rarity. However, many preoperative assessments may reveal it. For example, abdominal ultrasound to detect gallstones, as is conducted routinely in many centres, will point to *SIT*.^{9,10,13} Chest auscultation or chest

radiographs may reveal dextrocardia.^{12 13 32} Findings in electrocardiograms such as poor or reverse R-wave progression in the chest leads or predominantly negative complexes in lead I should raise suspicion of *SIT*.¹² When *SIT* is suspected, the diagnosis can be confirmed with chest radiography or ultrasonography.³³ CT or MRI can be used where anatomic questions remain.³³ In this case, the patient had known Kartagener syndrome since childhood.

The preoperative diagnosis of *SIT* can help to manage the risk of pulmonary, cardiac or other complications associated with the disease. Patients with *SIT* may have other developmental malformations,²² such as respiratory (bronchiectasis or congenital absence of one lung), cardiovascular (Tetralogy of Fallot) or digestive system (biliary atresia or small bowel atresia) anomalies.^{7 34} Patients should be investigated carefully for any associated anomalies.²² The literature recommends imaging techniques such as CT scan or MRI of the thorax and abdomen,^{17 19 30} abdominal ultrasounds^{5 9 10 13} or echocardiography.^{23 30} Endoscopy can identify hiatal hernias, abnormal appearances of the stomach or duodenal pathologies.^{16 30 32} Pulmonary function analysis is recommended to screen for conditions such as bronchiectasis.^{16 23 30} A multidisciplinary approach is needed to manage any anomalies. In this case, a respiratory physician informed the medical and surgical team about how to reduce the risk of intraoperative and postoperative complications that may have arisen from the patient's bronchiectasis.

Preoperative awareness of *SIT* allows the surgical team to prepare for the technical and nontechnical challenges of the surgery.^{22 24} For LSG, the entire operation is performed in a mirror-image fashion.^{22 23 30–32} Technical aspects such as the positioning of the team, trocar placements, and insertion of instruments must be planned carefully.^{22 23 30–32} In terms of non-technical challenges, the surgeons should be aware of difficulties in overcoming cognitive dissonance associated with the anomalous anatomy. Surgeons in the literature report that they learnt to adapt quickly and to perform the operation in reverse from usual.^{14 20 21 24} Wittgrove *et al*²¹ recommend auditory prompting to remind one another of the patient's anatomical differences and to over-ride automatic movements. In this case, the surgeons cross-checked at each stage of the operation to help overcome their cognitive dissonance.

In summary, increasing numbers of obese patients are predicted to elect bariatric surgery as a long-term treatment option.³⁵ Accordingly, patients with *SIT* and Kartagener syndrome can be expected to seek LSG and may be reassured that this procedure can be performed safely. This case report supports the literature

showing that LSG can proceed in patients with *SIT* or Kartagener syndrome, although with some technical and cognitive adjustments. The report suggests ways to reduce the risk of complications. This case report can help to inform future surgeons about how to manage similar patients.

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Learning points

- A multidisciplinary approach is important in bariatric surgery and may require further interdisciplinary collaboration in patients with Kartagener syndrome.
- Laparoscopic bariatric surgeries are technically demanding procedures in patients with *situs inversus totalis* (*SIT*), due to significant deviation from normal anatomy.
- This report presents the only identified case in the literature of multiple-incision laparoscopic sleeve gastrectomy performed on a Kartagener syndrome patient.
- Laparoscopic sleeve gastrectomy in patients with *SIT* can be safely performed by an experienced surgeon utilising appropriate changes in the operative technique and the positioning of the surgical team.

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