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Patient: Final Diagnosis: Symptoms: Medication: Clinical Procedure: Specialty:	Male, 35 Low-grade appendiceal m Abdominal pain — Single port laparoscopic c Surgery	·	ith hyperthermic intraperitoneal chemotherapy
Objective:	Unusual or unexpected effo	ect of treatment	
Background:	Cytoreductive surgery with hyperthermic intraperitoneal chemotherapy is one of the treatment options in low-grade appendiceal mucinous neoplasm with peritoneal dissemination. The minimal invasive surgery ap- proach was introduced to the field after years of traditional open technique. Multi-port laparoscopic and ro- botic techniques were reported with good short-term outcomes in very selected patients with low PCI scores. We describe here the first single-port laparoscopic approach of cytoreductive surgery and hyperthermic intra- peritoneal chemotherapy.		
Case Report:	We present a case of low-grade appendiceal mucinous neoplasm with peritoneal dissemination, in which single-port laparoscopic approach of cytoreductive surgery and hyperthermic intraperitoneal chemotherapy was completed through a 5.5-cm incision. A 35-year-old man with no medical illness underwent laparoscopic appendectomy for acute appendicitis 3 months earlier. Postoperative surgical pathology reported a low-grade appendiceal mucinous neoplasm with positive margin. After complete assessment and Tumor Board discussion, the patient was scheduled for cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. A single gel port access was inserted through a 5.5-cm peri-umbilical incision. The Peritoneal Cancer Index score was 4, and the decision was made to proceed with partial cecectomy, omentectomy, peritonectomy, and hyperthermic intraperitoneal chemotherapy with the Sugarbaker mitomycin C-based regimen. Postoperative care was carried out following the Enhanced Recovery After Surgery protocol. The patient was discharged on day 3 with-		
Conclusions:	out any complications. A single-port laparoscopic approach in cytoreductive surgery with hyperthermic intraperitoneal chemothera- py is safe and feasible in very selected patients, and has the advantage of direct visualization and palpation through the incision to determine more accurate Peritoneal Cancer Index assessment in comparison to other MIS approaches. Nevertheless, additional prospective studies are needed.		
MeSH Keywords:	Adenocarcinoma, Mucinous • Injections, Intraperitoneal • Laparoscopy • Surgical Procedures, Minimally Invasive		
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Background

Peritoneal surface malignancy (PSM) is defined as a group of tumors arising from abdominal organs, which can lead to peritoneal carcinomatosis (PC) [1]. Since its origin in the 1990s, cytoreductive surgery (CRS) with hyperthermic intraperitoneal chemotherapy (HIPEC) has evolved immensely and opened a new and promising way of treating incurable and palliative cases of PC [1]. The main aim of CRS is to debulk all visible parts of the tumor. This can be done by direct resection of tumor parts or by complete removal of the involved organ in order to achieve complete debulking as well as HIPEC to eradicate the microscopic disease [2].

In recent years, different minimal invasive methods for CRS-HIPEC have been described, including using robotics and laparoscopic hand-assisted techniques [3–5]. Here, we describe what we believe to be the first CRS and HIPEC performed through a single-port laparoscopic (SPL) approach.

Case Report

A 35-year-old man, not known to have any medical illnesses, presented with a history of acute appendicitis 3 months earlier, for which he underwent an uneventful laparoscopic appendectomy. However, the appendix was perforated and mucin was noted all over the specimen in the extraction bag. Upon reviewing his surgical pathology, a low-grade appendiceal mucinous neoplasm (LAMN) was shown with extra-appendiceal mucin at the resection margin and acellular mucin invading the visceral peritoneum. The proximal resection margin was involved; the mesenteric resection margin contained acellular mucin, and acute appendicitis was also identified. Investigations, including tumor markers and computerized tomography (CT) scans, were done. CEA level was 1.160 ng/mL. CA 19-9 was sent but the tracer reagent was not available. CT chest, abdomen, and pelvis reported unremarkable findings with no obvious disease. CRS with HIPEC was planned after Tumor Board discussion. Informed consent was obtained from the patient, and this report was prepared after approval by our institutional Ethics Committee.

Under general anesthesia and through a 5.5-cm peri-umbilical incision (Figure 1), which was deepened to reach the intra-abdominal cavity, a single gel port was inserted (Figure 2). Full exploration of the abdomen was completed, including direct inspection by the laparoscope camera, direct visualization through the single incision, and hand palpation of the retrohepatic space. The intraoperative findings included tumor deposits in the rectovesical pouch and right upper-quadrant abdominal wall, which were dissected and cleared. Mucin was also noted at the base of the appendix. The Peritoneal Cancer



Figure 1. A 5.5-cm peri-umbilical incision was made to introduce the port.



Figure 2. The single gel port was used to explore the abdomen with one 10-mm port and two 5-mm ports.

Index (PCI) [6] was 4. The right lower quadrant scored 2 for disease at the base of the cecum. There was one nodule in the rectovesical pouch and another nodule in the right upper quadrant, and both were scored 1. All the lesions were identified by the camera scope and palpation. Partial cecectomy and omentectomy were performed. In addition, HIPEC was done using the Sugarbaker mitomycin C-based regimen with mitomycin C and doxorubicin for intraperitoneal solution; both were in 2L 1.5 dextrose at a dose of 15 mg/m² and 42°C. Simultaneously, intravenous infusion of 5-flurourcil (400 mg/m²) and leucovorin (20 mg/m²) was started for 90 min [7].

The surgery went smoothly, without any complications during or after the operation. The Enhanced Recovery After Surgery (ERAS) protocol was followed. The patient resumed oral intake on the night of the procedure and was discharged home 3 days after the surgery. There were no complications during the 30-day follow-up. Postoperative surgical pathology reported a positive focus of low-grade mucinous neoplasm at the appendicular stump, with negative margin, including the cecum wall. There was 1 reactive lymph node negative for malignancy. The 2 peritoneal nodules resected in the right upper and lower quadrants were negative for malignancy.

Discussion

Tumors of appendicular origin are rare. They are estimated to occur in less than 2% of all patients who undergo an appendectomy, and 0.4% of all gastrointestinal tract malignancies [8]. Most cases present as acute appendicitis, and less frequently are incidentally found intra-operatively during surgery for other abdominal diseases [8]. Adenocarcinoma of the appendix is subdivided into colonic and cystic types based on the growth pattern. The colonic type arises from tubular or tubulovillous adenomas, and it mimics the course of adenocarcinoma of colon origin, while the cystic type results as a progression of a cystadenoma on the base of a villus adenoma [8,9]. Mucinous appendiceal neoplasms (MANs) are of the cystic type, with mucin-producing cells. MANs are the most common cause of pseudomyxoma peritonei (PMP), which present in about 20% of patients with MANs. PMP is a silent disease with no symptoms at the beginning. As the disease progresses, patients can present with nonspecific symptoms such as vague abdominal pain, distension, or hernia related directly to the accumulation of mucin [1,10].

In 2016, the Peritoneal Surface Oncology Group International classified mucinous neoplasms into 4 types: LAMN, high-grade appendiceal mucinous neoplasm (HAMN), mucinous adenocarcinoma, and poorly differentiated mucinous adenocarcinoma. As the diagnosis in our case, LAMN is defined as low-grade cytologic atypia with extension beyond the mucosa, but without any wall invasion [1]. Our case was a LAMN with acellular mucin, with better prognosis compared to other types of mucinous neoplasms.

In a retrospective, multi-center study, Chua et al. [11] evaluated the outcome and long-term survival rate in 1559 LAMN patients who underwent CRS-HIPEC. PCI remained the most significant prognostic factor, with 5-year and 10-year survival rates of 73% and 93% for patients with PCI of more than 31 and less than 10, respectively. Moreover, the completeness of cytoreductive resection has a strong influence on the survival rate, with 5-year and 10-year survival rates of 91% and 81%, respectively, in complete resection. When compared with incomplete cytoreductive resection and residual disease more than 2.5 mm, the 5-year and 10-year survival rates were 33% and 10%, respectively [11]. In our case, we did a complete abdominal exploration and PCI assessment with direct inspection by the laparoscopic camera, direct visualization, and palpation of bilateral subphrenic area through the single incision. PCI was 4. Given that the margin was positive in the first surgery, the preoperative CT scan assessment was negative for remnant disease and we were satisfied with our PCI assessment intra-operatively. We elected to complete CRS and HIPEC using SPL technique, which was achieved with complete resection.

The extended implementation of laparoscopy and MIS in the last 2 decades has reflected positively on surgeon experience and skills and has allowed for more MIS procedures. In contrast to the traditional open technique with a midline laparotomy incision, MIS in CRS-HIPEC has gained more attention in the last few years, with multiple published works describing its significance and benefits [3-5]. It is essential to mention that the main limitation of MIS in CRS is performing a complete abdominal exploration to determine an accurate PCI; more specifically, assessing the 2 areas behind the liver and spleen [12]. Passot and colleagues [4] conducted a prospective multicenter study evaluating the estimation of PCI by laparoscopy versus open technique in recurrence of peritoneal metastasis from colorectal cancer. They found that laparoscopy assessment failed to identify peritoneal deposit in 18% of the patients compared with 0% using the laparotomy approach.

Few studies and case reports have been published on the MIS approach in CRS-HIPEC with a curative intent. Salti et al. [3] published a report on 11 cases of PSM with a PCI of less than or equal to 10 managed by CRS-HIPEC using the hand-assisted laparoscopic approach. The short-term results and outcomes were better than with the traditional open technique. In another similar study, 8 patients with different peritoneal pathologies and a PCI of 10 or less underwent laparoscopic CRS-HIPEC, and the authors concluded that MIS is safe and feasible in the selected patients with PSMs and a low PCI [4]. Gabriel et al. [5] reported the first robotic-assisted CRS-HIPEC performed in a patient with residual disseminated peritoneal adenomucinosis secondary to LAMN and a PCI of 1; they reported improvement of the short-term outcome, with a short postoperative length of stay (4 days). Here, we report what we believe to be the first CRS-HIPEC performed through a SPL approach, showing the advantage of direct visualization and hand palpation of the bilateral subphrenic area for more accurate PCI assessment in comparison to the other MIS approaches. Additionally, we helped minimize the size and number of incision sites compared to traditional laparoscopy, which could minimize the risk of port-site metastasis.

The minimal invasive approach that we used and the ERAS protocol that we followed helped the patient's postoperative recovery course, including the avoidance of nasogastric tube insertion, early removal of the Foley catheter, immediate mobilization, early resumption of oral intake, and adequate analgesia. This is in addition to the short postoperative length of stay, which was 3 days.

We showed that the single-port approach is safe and feasible, with a positive short-term outcome, better patient recovery, and a shorter hospital stay in very selected patients with favorable condition and limited disease, as determined by preoperative histopathology and radiological study. However, the traditional open technique remains the standard of care, with the most accurate PCI assessment in comparison to all MIS approaches [4]. Further studies are needed to better evaluate the single-port approach in CRS-HIPEC and to assess the short-term and long-term advantages.

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Conclusions

Single-port laparoscopic CRS-HIPEC is safe and feasible in very selected patients. It has the advantages of direct visualization and palpation through the incision to determine more accurate PCI assessment in comparison to other MIS approaches. Nevertheless, additional prospective studies are needed.

Conflicts of interest

None.

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