



# EPIGASTRIC PORT SITE METASTASIS IN AN UNKNOWN PRIMARY: A RARE CASE REPORT

D.S. PRUTHI, P. NAGPAL, M. PANDEY

Department of Radiation Oncology, Action Cancer Hospital, New Delhi, India

**Abstract – Objective:** The advent of laparoscopic oncologic surgery has brought with it the peculiar issue of port-site metastasis (PSM). The most common source of primary site for PSM is from gall bladder, ovarian and pancreas in which some laparoscopic procedure has been done. PSM in a case of unknown primary is very rare.

**Case report:** Here in we present a case of a 71-year-old female who had underwent laparoscopic cholecystectomy for cholelithiasis 2 years ago. There was no evidence of malignancy at that time. The patient presented after 2 years of the surgery with a mass at the epigastric port site region which was confirmed to be metastatic adenocarcinoma after histopathology and immunohistochemistry. The site of primary tumor could not be identified even after thorough investigations.

**Results:** The patient underwent wide local excision of the mass followed by adjuvant radiotherapy and chemotherapy for the same. To our knowledge this is the 3rd case reported in literature in which PSM has been found with primary unknown in post cholecystectomy for chronic cholecystitis.

**Conclusions:** The treating physician should be aware that there is a possibility of development of PSM after a latency period ranging from a few months to years. The most common method to avoid PSM is to prevent intraoperative spillage.

**KEYWORDS:** Port site metastasis, Unknown primary, Adenocarcinoma.

Despite various advantages of a minimally invasive approach, oncological safety of laparoscopy has been a point of debate due to occurrence of Port Site Metastasis (PSM) and tumor seeding. Port Site Metastasis (PSM) is defined as recurrent cancerous lesions developing locally in the abdominal wall within the scar tissue at one or more trocar sites<sup>1</sup>. Port site metastasis has been previously reported in cases of laparoscopic oncologic surgery, video-assisted thoracoscopic surgery and robot-assisted oncologic surgery. The estimated incidence of port-site metastasis in all patients undergoing laparoscopic surgery for malignant disease is approximately 1-2%<sup>1</sup>. Laparoscopic assisted surgeries are most commonly done in gastrointestinal, gynecology and uro-oncology.

According to three large-scale, multicentre, and randomized controlled studies (COST, COLOR, and UK MRC CLASICC) reported in 2007, 2009, and 2010, respectively, the incidence of port-site metastasis was 0.9% (2/435), 1.3% (7/534) and 1.7% (9/526) respectively. The incidence was higher in patients with T4 stage cancer<sup>2</sup>. In a review of 12 years of laparoscopic surgeries for gynecologic malignancies, they found port-site recurrences in 0.97% of cases and majority of cases occurred within 1 year<sup>3</sup>. Of the 1288 patients, seven developed laparoscopic port-site metastasis, and seven developed implants at the site of intraperitoneal catheter ports. The incidence of PSM was noticed in surgeries for advanced or recurrent cases like carcinomatosis<sup>3</sup>. In urologic oncologic laparoscop-



This work is licensed under a [Creative Commons Attribution-NonCommercial-ShareAlike 4.0 International License](https://creativecommons.org/licenses/by-nc-sa/4.0/)

DOI: 10.32113/wcrj\_20229\_2401



ic studies, the incidence has been reported from 0.09% to 0.73%<sup>4</sup>. It is believed that laparoscopic procedures can affect the prognosis of the disease by increasing the risk of port site or peritoneal seedling by upstaging the disease<sup>5</sup>. Its etiology is uncertain, and it is deemed to be multifactorial. The mechanism is unknown but there are several hypotheses: hematogenous spread, direct contamination, effects of pneumoperitoneum, and aerosolization of tumor cells<sup>6</sup>. Herein we present a case of epigastric port site adenocarcinoma in a 71-year-old female with no evidence of primary disease.

### CASE PRESENTATION

A 71-year-old female with comorbidities of diabetes and hypertension presented with complaints of pain and swelling in the upper abdomen for 2 months duration. She gives history of undergoing laparoscopic cholecystectomy for cholelithiasis 2 years ago. Magnetic resonance cholangiopancreatography (MRCP) was done at that time and had revealed cholelithiasis. The post-operative histopathology was acute on chronic cholecystitis with cholelithiasis.

Two years after her surgery, the patient developed pain abdomen, which was gradually progressive, dull aching, non-radiating, and non-referred. She then presented to the hospital. On examination there was a hard epigastric mass palpable beneath the previous port site wound with tenderness. It was not differentiated separately from the xiphisternum and not fixed to overlying skin. A contrast CT scan of the abdomen showed a well-defined

isodense mass lesion seen in the deep planes of the anterior abdominal wall measuring 2.4 x 6.1 x 9.2 cm having speculated margins with few chunky calcifications. On post contrast images it is showing heterogeneous enhancement. Posteriorly it is invading the omentum. Biopsy revealed metastatic deposits of adenocarcinoma. Upper GI endoscopy and colonoscopy were normal. Mammography was also normal. Blood investigation revealed a normal complete blood count, liver function tests and kidney function tests. CA19.9 was 5.88 U/mL while CEA was 0.95 ng/mL. Serum amylase was slightly raised being 50 IU/L.

PET CT scan (Figure 1) showed intensely FDG avid (SUV max 20) heterogeneously enhancing intramuscular soft tissue lesion involving lower chest and upper abdominal wall with a few specks of calcification within the perilesional thickening and stranding with mild intraabdominal extension. It measures 2.8 (AP) x 3.8 (TR) x 8.2 (CC) cm in dimensions. Mildly FDG avid (SUVmax 3.7) subcentimetric aortocaval lymph node seen. There is presence of few FDG avid (SUVmax 16.2) left level II and III cervical lymph nodes largest measuring 1.1 x 0.9 cm. No other lesions identified anywhere else in the body.

An ultrasound guided FNAC from the left cervical lymph node region showed reactive lymphadenitis and did not reveal any evidence of malignancy.

The patient then underwent a diagnostic laparoscopy with wide local excision of mass. Hard mass was present below the port side. Wide local excision was done, which included the muscle and falciform ligaments.

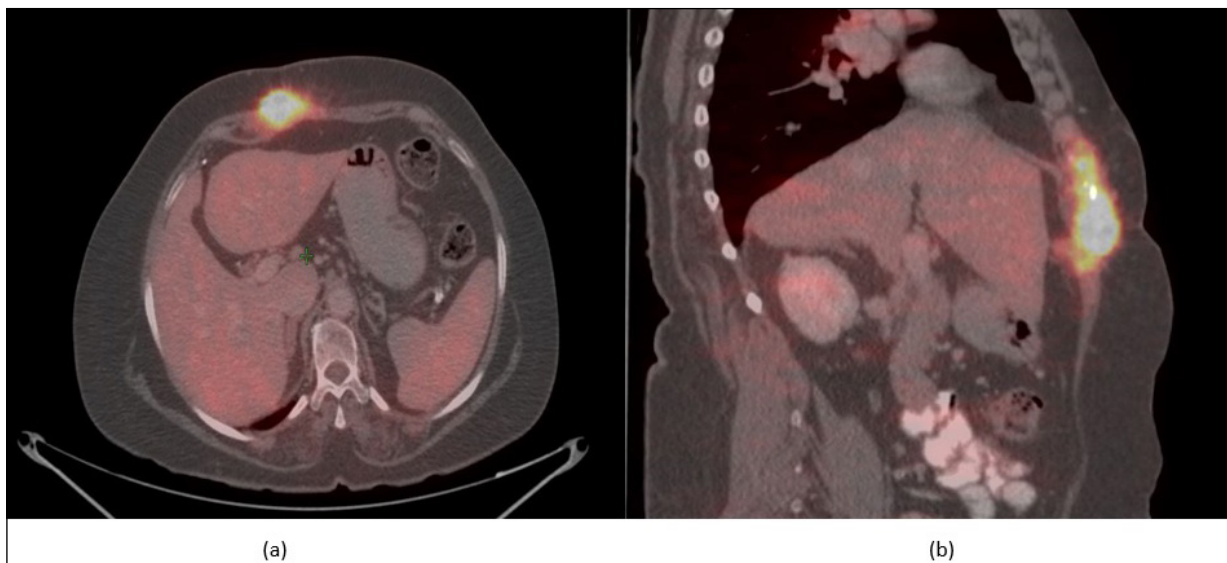


Fig. 1. Sections of PET CT scan of the patient showing epigastric nodule (a) axial view and (b) sagittal view.

On histopathology, irregular, grey-white gritty mass 8 x 6 x 4 cm was seen. On microscopy, presence of diffusely infiltrating tumor predominantly based in the subcutaneous plane and arranged in the form of irregularly placed glands, nests and single cell infiltrate embedded in inflammatory rich desmoplastic stroma suggestive of adenocarcinoma. Tumor is extensively infiltrating the skeletal muscle bundles. Foci of ossification are seen within the tumor. Resected base was involved by tumor which was seen to reach the peritoneal surface. Tumor was focally reaching up to the deeper dermis. Rest of the margins were free. On immunohistochemistry the tumor cells were positive for CK7 and CK20 and PAX8 were both negative hence confirming the diagnosis of metastatic adenocarcinoma.

The patient was then treated with post-operative radiotherapy to the port site tumor bed with biological equivalent dose of 60Gy using 6 Mega Voltage (MV) photon beams. The further course of treatment is adjuvant chemotherapy (gemcitabine based).

## DISCUSSION

Laparoscopic surgeries are associated with several desirable advantages such as lesser pain, quicker recovery and shorter hospitalization making them a widely adopted procedure for patients with intra-abdominal disease<sup>7</sup>. It has also become a routine staging procedure for various malignancies like gastric, ovarian etc. However, one shortcoming of such procedures is the development of PSM. The incidence of PSM is variable in literature from 0.71% to 21%<sup>8,9</sup>. In fact, in one rare scenario, PSM occurred in a patient undergoing staging or diagnostic laparoscopy where no tumor had been identified<sup>10</sup>.

Its etiology is unknown and can involve multiple factors. Direct implantation of tumor cells as tumor contamination on operating instruments has been reported<sup>10</sup>. In this regard, the operating port is most commonly involved<sup>11</sup>. The operating port encounters higher contamination with tumor cells. Injury to the peritoneum and abdominal wall at the trocar sites have been shown to increase the chances of tumor implantation. In other theories, hematogenous spread has been implicated<sup>12</sup>. Another theory states that the type of gas used, the pressure and duration of surgery can also be responsible for tumor seedling. This is because the peritoneum gets injured due to introduction of trocar. Carbon dioxide irritates the peritoneum causing inflammatory changes and acidosis which may cause implantation<sup>9</sup>. This theory was highlighted by Wu et al<sup>13</sup> in which they suggest-

ed that pneumoperitoneum at 10 mmHg increases wound implantation in cases of intra-abdominal tumor spillage. It also depends on the surgeon's technique and tumor handling<sup>14</sup>. In extremely rare instances aerosolisation of tumor cells have been suggested<sup>15</sup>. This shows that the etiology of PSM can be multifactorial and to pinpoint one particular reason is difficult.

There have been several methods which have been proposed to reduce inoculation of malignant cells like using endobag while delivering the specimen, excision of port site, use of helium gas or gasless laparoscopy and avoid manipulation of tumor<sup>16</sup>. Some recommend the use of intraperitoneal cytotoxic drugs<sup>17</sup>. In short, the best way to prevent PSM is to avoid direct tumor handling and strictly adhere to the principles of laparoscopic oncology principles. When there is a delay between the laparoscopic surgery and development of PSM, settling down of unknown circulating malignant cells at the injured site is thought to be the causative factor. In our patient, despite full body investigations, source of primary malignancy could not be found, and this might have been a manifestation of an occult primary and the above-mentioned reasons might have resulted in the secondary site of metastasis after 2 years.

Two similar case reports were found after doing a literature search. In one study, a 75-year-old male had undergone laparoscopic cholecystectomy for cholelithiasis and there was no evidence of any malignancy in the specimen. However, 11 months later, he developed subcutaneous nodules on two sites: port site at right axillary line (vacuum drain) and sub umbilical region. The histopathology showed metastatic adenocarcinoma. The patient underwent wide local excision of both the nodules. However, 6 months later the patient developed recurrence in both the sites and again underwent surgery followed by radiotherapy. In another study, a 45-year-old woman presented with a single epigastric PSM which was found to be papillary adenocarcinoma. The patient had a history of undergoing laparoscopic cholecystectomy for cholelithiasis 28 months prior to current presentation. The patient underwent wide local excision followed by chemotherapy<sup>18</sup>. In the other study, 45-year-old female underwent laparoscopic cholecystectomy which was found to be chronic cholecystitis on histopathology. After 28 months the patient presented with an epigastric mass and underwent surgery which showed papillary adenocarcinoma. The site of primary tumor could not be identified despite all investigations. The patient was started on adjuvant chemotherapy. This is an example of delayed presentation of a port site metastasis similar to our case<sup>19</sup>.



Our case of port site metastasis with unknown primary presented here is similar to the ones reported above as all the patients had undergone laparoscopic cholecystectomy for cholelithiasis with no evidence of primary disease. To our knowledge this is the 3<sup>rd</sup> case report in literature in which there is presence of PSM with unknown primary in patients who had undergone laparoscopic cholecystectomy for cholecystitis.

Port site metastasis are usually seen from an unsuspected primary in the abdomen mostly commonly from the gall bladder, pancreas, or ovary. The incidence of PSM after laparoscopic cholecystectomy of unsuspected gall bladder carcinoma is 0.5-1% of the performed LC's<sup>20</sup>.

## CONCLUSIONS

The treating physician should be aware that there is a possibility of development of PSM after a latency period ranging from a few months to years. The most common method to avoid PSM is to prevent intraoperative spillage. Regular follow up of the patients is very important. Treatment of such cases should be surgery with clear margins followed by post-operative radiotherapy for local control and adjuvant chemotherapy for systemic control.

## AUTHOR CONTRIBUTION:

Deep Shankar Pruthi: Collection of data, analysis and writing of manuscript.

Puneet Nagpal: Collection of data and proof reading.

Manish Pandey: Proof reading and supervision.

## CONFLICT OF INTEREST:

The authors have no conflict of interest to declare.

## FUNDING:

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

## ETHICAL APPROVAL:

This article was submitted and approved by the responsible Ethics Committee.

## INFORMED CONSENT:

Informed consent was obtained from the participant included in the study.

## REFERENCES

1. Hung GU, Hsu HK, Kao CH, Chen KY, Chiu JS. Asymptomatic port-site metastasis following video-assisted thoracoscopic surgery detected by FDG-PET/CT. *Clin Nucl Med* 2010; 35: 552-553.
2. Wang YY, Qian ZY, Jin WW, Zhao ZK, Zhang W, Mou YP. Surgical Treatment of Port-Site Metastases After Laparoscopic Radical Resection of Gastrointestinal Tumors. *J Laparoendosc Adv Surg Tech A* 2020; 30: 1090-1094.
3. Abu-Rustum NR, Rhee EH, Chi DS, Sonoda Y, Gemignani M, Barakat RR. Subcutaneous tumor implantation after laparoscopic procedures in women with malignant disease. *Obstet Gynecol* 2004; 103: 480-487.
4. Micali S, Celia A, Bove P, De Stefani S, Sighinolfi MC, Kavoussi LR, Bianchi G. Tumor seeding in urological laparoscopy: An international survey. *J Urol* 2004; 171: 2151-2154.
5. Wibbenmeyer LA, Wade TP, Chen RC, Meyer RC, Turgeon RP, Andrus CH. Laparoscopic cholecystectomy can disseminate in situ carcinoma of the gallbladder. *J Am Coll Surg* 1995; 181: 504-510.
6. Gao KJ, Yan ZL, Yu Y, Guo LQ, Hang C, Yang JB, Zhang MC. Port-site metastasis of unsuspected gallbladder carcinoma with ossification after laparoscopic cholecystectomy: A case report. *World J Clin Cases* 2020; 8: 5729-5736.
7. Toouli J, Cox MR. Minimal access surgery of the gastrointestinal tract. *Aust N Z Surg* 1995; 65: 525-532.
8. Ziprin P, Ridgway PF, Peck DH, Darzi AW. The theories and realities of port-site metastases: a critical appraisal. *J Am Coll Surg* 2002; 195: 395-408.
9. Berends FJ, Kazemier G, Bonjer HJ, Lange JF. Subcutaneous metastases after laparoscopic colectomy. *Lancet* 1994; 344: 58.
10. Bouvy ND, Marquet RL, Jeekel H, Bonjer HJ. Impact of gas(less) laparoscopy and laparotomy on peritoneal tumor growth and abdominal wall metastases. *Ann Surg* 1996; 224: 694-701.
11. Curet MJ, Putrakul K, Pitcher DE, Zucker KA. Laparoscopically assisted colon resection for colon carcinoma. *Surg Endosc* 2000; 14: 1062-6.
12. Iwanaka T, Arya G, Ziegler MM. Mechanism and prevention of port-site tumor recurrence after laparoscopy in a murine model. *J Pediatr Surg* 1998; 33: 457-461.
13. Wu JS, Brasfield EB, Guo LW, Ruiz M, Connett JM, Philpott GW, Jones DB, Fleshman JW. Implantation of colon cancer at trocar sites is increased by low pressure pneumoperitoneum. *Surgery* 1997; 122: 1-7.
14. Mathew G, Watson DI, Rofe AM, Baigrie CF, Ellis T, Jamieson GG. Wound metastases following laparoscopic and open surgery for abdominal cancer in a rat model. *Br J Surg* 1996; 83: 1087-1090.
15. Tseng LN, Berends FJ, Wittich P, Wittich P, Bouvy ND, Marquet RL, Kazemier G, Bonjer HJ. Port-site metastases. Impact of local tissue trauma and gas leakage. *Surg Endosc* 1998; 12: 1377-80.
16. Schneider C, Jung A, Reymond MA, Tannapfel A, Balli J, Franklin ME, Hohenberger W, Köckerling F. Efficacy of surgical measures in preventing port-site recurrences in a porcine model. *Surg Endosc* 2001; 15: 121-5.
17. Balli JE, Franklin ME, Almeida JA, Glass JL, Diaz JA, Reymond M. How to prevent port-site metastases in laparoscopic colorectal surgery. *Surg Endosc* 2000; 14: 1034-1036.
18. Polychronidis A, Tsaroucha AK, Perente S, Giatromanolaki A, Koukourakis M, Simopoulos C. Port-site metastasis of extrahepatic bile duct carcinoma after laparoscopic cholecystectomy without evidence of a primary tumour. *Acta Chir Belg* 2008; 108: 768-770.
19. Rao S, Rathod A, Kamble A, Gupta D. Delayed presentation of port-site metastasis from an unknown gastrointestinal malignancy following laparoscopic cholecystectomy. *Singapore Med J* 2014; 55: e73-e76.
20. Nakagawa S, Tada T, Furukawa H, Abe M, Hatakeyama K. Late-type recurrence at the port site of unexpected gallbladder carcinoma after a laparoscopic cholecystectomy: report of a case. *Surg Today* 2000; 30: 853-855.