Small intestinal metastases from esophageal carcinoma presenting as a perforation: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Small intestinal metastasis from esophageal carcinoma is rare. We report a case of small intestinal metastases from esophageal carcinoma presenting as a perforation and discuss the aetiology with other cases of small intestinal metastasis from esophageal carcinoma reported in previous literature.

PRESENTATION: An 86-year-old man presented with fever and coughing. He had choked while eating and had history of weight loss. He was diagnosed with aspiration pneumonia. Two days after the admission, he complained of abdominal pain. Physical examination revealed guarding and rebound tenderness in the upper abdomen. A contrast computed tomography of the abdomen showed ascites, free air, and irregular thickness of the small intestinal walls. Small intestinal perforation was noted, and surgical resection of the small intestine was performed. The pathological findings of the resected small intestine revealed ulcers with squamous cell carcinoma, and upper gastrointestinal endoscopy demonstrated esophageal tumour, whose biopsy revealed squamous cell carcinoma. A diagnosis of small intestinal metastases from esophageal carcinoma was made, but the patient died one month after the diagnosis.

DISCUSSION: Most cases found in the literature of esophageal tumour involve squamous cell carcinoma with male patients, and specific symptoms are divided into obstruction and perforation. All patients with small intestinal metastasis from esophageal carcinoma who survived were treated by a combination of resection and radiation and/or chemotherapy; thus, immediate treatments seem essential to improve the prognosis.

CONCLUSION: Physicians should keep in mind the possibility of small intestinal metastasis when patients with a history of esophageal cancer have abdominal symptoms.

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1. Introduction

Metastatic involvement of the intestinal tract from extra-abdominal sites is uncommon [1]. In esophageal carcinoma, liver and lungs are the most common sites for metastases, and small intestinal metastasis from esophageal carcinoma has rarely been reported in the previous literature [2–4]. We report a rare case of small intestinal metastases from esophageal carcinoma presenting as a perforation and discuss the aetiology with other cases of small intestinal metastasis from the esophageal carcinoma reported in previous literature. The work in this case has been reported in line with the surgical case report (SCARE) criteria [5].

2. Case presentation

An 86-year-old Japanese man presented with a history of fever and cough. He had sometimes been choked with even soft foods and had 5 kg of body weight loss for the last 3 months. He had a history of diabetes mellitus, chronic renal failure, dyslipidaemia, and dementia and was taking medications including insulin for these conditions. He had been smoking 20 cigarettes per day for 66 years and drinking a glass of sake per day for 60 years. His family history was unremarkable. On arrival, his blood pressure, pulse, body temperature, respiratory rate, and oxygen saturation were 153/56 mmHg, 100 beats/min, 38.5 °C, 16 breaths/min, and 98% under room air, respectively. A physical examination revealed coarse crackles on the bilateral lower lobes and no tenderness on the abdomen. Laboratory studies revealed findings of leucocytosis, an increased level of C-reactive protein (CRP), renal dysfunction, hyperglycaemia, and an increased level of glycated haemoglobin, which indicated uncontrolled diabetes mellitus.
Table 1
Laboratory findings on the admission day.

| Test   | Value       | Test   | Value        | Test   | Value     |
|--------|-------------|--------|--------------|--------|-----------|
| WBC    | 13.8 10^9/mm^3 | CPK    | 39 IU/L | Na    | 134 mEq/L |
| Neu    | 91.1 %      | T-BIL  | 0.9 mg/dL | K     | 4.5 mEq/L |
| Lym    | 5.6 %       | AST    | 20 IU/L | Cl    | 103 mEq/L |
| Mono   | 3.0 %       | ALT    | 13 IU/L | Ca    | 10.1 mg/dL|
| RBC    | 4.55 10^12/μL | LDH    | 266 IU/L | Mg    | 2.1 mg/dL |
| Hb     | 13.0 g/dL   | γGTP   | 19 IU/L | IP    | 2.1 mg/dL |
| Ht     | 38.7 %      | TP     | 7.9 g/dL | Glu   | 289 mg/dL |
| MCV    | 85.1 fl     | ALB    | 3.2 g/dL | HbA1c | 8.2 %     |
| PLT    | 202 10^3/μL | BUN    | 32.1 mg/dL | CEA   | 7.6 ng/mL |
| CRE    | 1.34 mg/dL  | CYFRA  | 6.9 ng/mL  |
| CRP    | 3.4 mg/dL   | SCC antibody | 6.6 ng/mL |

Fig. 1. a) A marked thickening of the middle intra-thoracic oesophageal wall (red arrow). b) A bilateral infiltration and a mass (blue arrow), 10 x 10 mm in size, in the right S9 lesion.

(a) A computed tomography (CT) scan of the chest showed marked thickening of the middle intrathoracic oesophageal wall, bilateral infiltration, and a mass, 10 x 10 mm in size, in the right segment 9 (S9) lesion (Fig. 1). Thus, we diagnosed the patient with aspiration pneumonia and suspected lung and oesophageal carcinoma and started broad-spectrum antibiotics. Two days after the admission, the patient suddenly complained of abdominal pain. Physical examination revealed muscular defence and rebound tenderness in the upper abdomen. A contrast CT scan of the abdomen showed ascites, free air, irregular thickness of the small intestinal walls, and mesenteric lymphadenopathy (Fig. 2). Emergency laparotomy was performed. Intraoperative findings showed that a non-perforated ulcer with a submucosal nodule approximately 80 cm distal from the ligament of Treitz and a perforated ulcer approximately 110 cm distal from the ligament of Treitz (Fig. 3).

A diagnosis of small intestinal perforation was made, and surgical resection of the small intestine 30 cm in length, end-to-end anastomosis, and saline lavage in the abdominal cavity were performed. Markers panel like p63 and cytokeratin 5/6 is highly sensitive and specific for distinguishing squamous cell carcinoma from adenocarcinoma, and immunohistochemical results of these two ulcerative lesions positive for both p63 and cytokeratin 5/6. Thus the pathological findings of the resected small intestine revealed ulcers with squamous cell carcinoma, which suggests the primary site of carcinoma is different since the other sites of the small intestine did not have any findings of malignancy and primary squamous cell carcinoma of the small intestine is extremely rare (Fig. 4a-e). On postoperative day 7, upper gastrointestinal endoscopy was performed, and ulcerative and localised type of oesophageal tumour as the macroscopic classification was
Table 2  
Case reports of small intestine metastasis from esophageal carcinoma in English literature.

| Case | Ref | Year | Author | Age | Sex | Primary esophageal carcinoma | Metastatic tumor of small intestine | Other metastasis | Treatment | Outcome after the metastasis (duration) |
|------|-----|------|--------|-----|-----|-------------------------------|-----------------------------------|----------------|-----------|----------------------------------------|
| 1    | 2   | 1983 | Wang   | 65  | M   | Lt SCC                        | Obstruction                      | None           | Res       | ND                                    |
| 2    | 5   | 1988 | Williams | 60  | M   | Mt SCC                        | Obstruction                      | None           | Res       | ND                                    |
| 3    | 3   | 1996 | Yamada | 56  | M   | Mt SCC                        | Obstruction                      | None           | Res       | ND                                    |
| 4    | 6   | 2005 | Neve   | 56  | M   | Lt SCC                        | Obstruction                      | None           | Res       | ND                                    |
| 5    | 8   | 2005 | Sreenarasimhaiah | 62 | M   | Lt SCC                        | Obstruction                      | None           | CRT       | ND                                    |
| 6    | 1   | 2005 | Lindenmann | 54 | M   | Mt SCC                        | Occasional found                 | None           | Res       | Alive (12 months)                     |
| 7    | 10  | 2005 | Arulraj | 52 | M   | ND SCC                        | CRT                              | None           | CRT       | ND                                    |
| 8    | 9   | 2009 | Daisari | 42 | M   | Lt AC                         | Obstruction                      | Lymph         | Mesenteric LN | ND                          |
| 9    | 11  | 2009 | Horio  | 72  | M   | Lt SCC                        | Obstruction                      | None           | Res       | Alive (9 months)                     |
| 10   | 12  | 2013 | Yamada | 69  | M   | Lt SCC                        | Obstruction                      | None           | Res       | Dead (6 months)                      |
| 11   | 4   | 2015 | Chino  | 71  | M   | Lt SCC                        | Obstruction                      | None           | Res       | Dead (9 months)                      |
| 12   | 13  | 2017 | Morinaga | 72 | M   | Mt SCC                        | Obstruction                      | Lymph         | Liver Bone | Alive (1 month)                     |
| 13   | -   | 2018 | Our case | 86 | M   | Mt SCC                        | None                            | Lymph         | Hemat     | Dead (1 month)                      |

Note: Ref=References, M=Male, Lt=Lower thoracic esophagus, Mt=Middle thoracic esophagus, ND=Not described, SCC=Squamous cell carcinoma, AC=Adeno carcinoma, Res=Resection of esophagus, CRT=Chemo-radiation-therapy, Chem=Chemotherapy, Rad=Radiation therapy, Lymph=Lymphogenous, Hemat=Hematogenous.

3. Discussion

Metastasis of esophageal carcinoma to the small intestine is extremely rare, and squamous cell carcinoma is more frequent than adenocarcinoma. The incidence of small intestinal metastasis from esophageal carcinoma is not well established, but it has been reported in previous English literature [1-4]. Our case is the second reported case of small intestinal metastasis from esophageal carcinoma in the literature.

Specific symptoms of small intestinal metastasis from esophageal carcinoma are not well documented. The most common symptom is abdominal pain, but other symptoms such as weight loss, nausea, and vomiting may also be present.

In our case, the patient presented with symptoms of intestinal obstruction, which was diagnosed on CT scans. The CT scans showed a mass in the small intestine, which was confirmed on further investigation with an upper gastrointestinal endoscopy.

The diagnosis of small intestinal metastasis from esophageal carcinoma can be challenging, and can be confirmed with a biopsy or surgical exploration. However, the correct diagnosis is often delayed due to the non-specific symptoms and the lack of awareness of this complication.

Fig. 2: Intraoperative findings showed the perforated surface of the small intestine. The patient was diagnosed with advanced esophageal squamous cell carcinoma, and the tumor was located in the lower esophagus. The patient underwent a subtotal esophagectomy and a jejunal interposition for reconstruction.

The patient experienced postoperative complications and died one month after the operation. The pathological examination revealed a 28 cm esophageal adenocarcinoma with lymph node involvement.

The presence of small intestinal metastasis from esophageal carcinoma is associated with advanced stage of the primary tumor, and the prognosis is generally poor due to the advanced disease at presentation. Therefore, early detection and adequate management of small intestinal metastasis from esophageal carcinoma are crucial for improving the prognosis.
the vertebral venous plexus, is also considered to be a possible mechanism of metastasis to the abdomen [4]. In our case, it is unlikely that peritoneal seeding during operation occurred because the resection of primary oesophageal cancer was not performed. However, lymphoid or haematogenous spreading cannot be identified. Other metastasis sites such as the lungs and liver are reported in the previous literature, which is consistent with the fact that the most common sites of the metastases from the oesophagus are the lungs and liver.

In the previous literature, 6 patients died within 3 years after the diagnosis of intestinal metastasis from oesophageal carcinoma, and 4 patients survived although the followed duration is within a year. The outcomes of the other 3 patients were not described. All patients with small intestinal metastasis from oesophageal carcinoma who survived were treated by a combination of resection and radiation and/or chemotherapy. These results may indicate that the combination of resection and chemoradiotherapy seems
to be essential to improve the prognosis, but a longer follow-up and accumulation of the cases are needed.

4. Conclusion

We report a rare case of intestinal metastases from oesophageal carcinoma and review the previous literature. All patients with small intestinal metastasis from oesophageal carcinoma who survived were treated by a combination of resection and radiation and/or chemotherapy; thus, immediate treatments seem essential to improve the prognosis. Physicians should be careful and keep in mind the possibility of small intestinal metastasis when patients with a history of oesophageal cancer have abdominal symptoms such as abdominal pain or vomiting.

Conflict of interest statement

None.

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Ethical approval

The ethical committee of our institution exempted the approval of this report.

Consent

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

Author contribution

Dr. Ryohei Ono, the first and corresponding author, drafted and finalised the manuscript. Dr. Hidemitsu Ogino and Dr. Yuto Igarashi performed the surgery. Other doctors, Jun Kawachi, Rai Shimoyama, Hiroyuki Kashiwagi, Naoko Isogai, Katsunori Miyake, Ryuta Fukai, Takaaki Murata, and Nobuaki Shinozaki have cooperated in this manuscript.

Registration of research studies

None.

Guarantor

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