



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

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



Ryohei Ono*, Hidemitsu Ogino, Jun Kawachi, Rai Shimoyama, Hiroyuki Kashiwagi, Naoko Isogai, Katsunori Miyake, Ryuta Fukai, Takaaki Murata, Yuto Igarashi, Nobuaki Shinozaki

Department of Surgery, Shonan Kamakura General Hospital, Japan

ARTICLE INFO

Article history:

Received 11 April 2018

Received in revised form 17 May 2018

Accepted 26 May 2018

Available online 5 June 2018

Keywords:

Small intestinal metastasis

Oesophageal carcinoma

Perforation

Aspiration pneumonia

Review of the literature

Case report

ABSTRACT

INTRODUCTION: Small intestinal metastasis from oesophageal carcinoma is rare. We report a case of small intestinal metastases from oesophageal carcinoma presenting as a perforation and discuss the aetiology with other cases of small intestinal metastasis from oesophageal 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 oesophageal tumour, whose biopsy revealed squamous cell carcinoma. A diagnosis of small intestinal metastases from oesophageal carcinoma was made, but the patient died one month after the diagnosis.

DISCUSSION: Most cases found in the literature of oesophageal tumour involve squamous cell carcinoma with male patients, and specific symptoms are divided into obstruction and perforation. 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.

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

© 2018 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

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

* Corresponding author at: Department of Surgery, Shonan Kamakura General Hospital, Okamoto1370-1, Kamakura City, Kanagawa, Japan.
E-mail address: ryohei.ono.0820@yahoo.co.jp (R. Ono).

Table 1
Laboratory findings on the admission day.

Complete Blood Count		Biochemistry			Coagulation		
WBC	13.8 $10^3/mm^3$	CPK	39 IU/L	Na	134 mEq/L	PT-%	77.2 %
Neu	91.1 %	T-BIL	0.9 mg/dL	K	4.5 mEq/L	PT-INR	1.13
Lym	5.6 %	AST	20 IU/L	Cl	103 mEq/L	APTT	26.8 sec
Mono	3.0 %	ALT	13 IU/L	Ca	10.1 mg/dL		
RBC	4.55 $10^6/\mu L$	LDH	266 IU/L	Mg	2.1 mg/dL		
Hb	13.0 g/dL	γ GTP	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 $10^3/\mu L$	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		

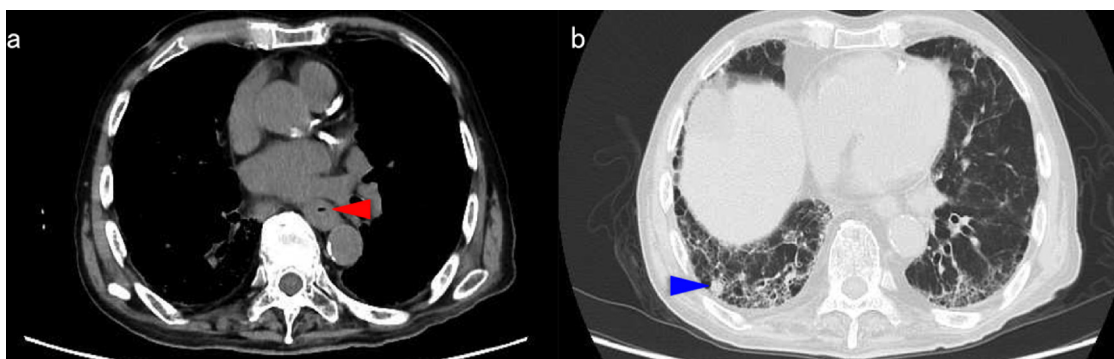


Fig. 1. a) A marked thickening of the middle intra-thoracic oesophageal wall (red arrow). b) A bilateral infiltration and a mass (blue arrow), 10 × 10 mm in size, in the right S9 lesion.

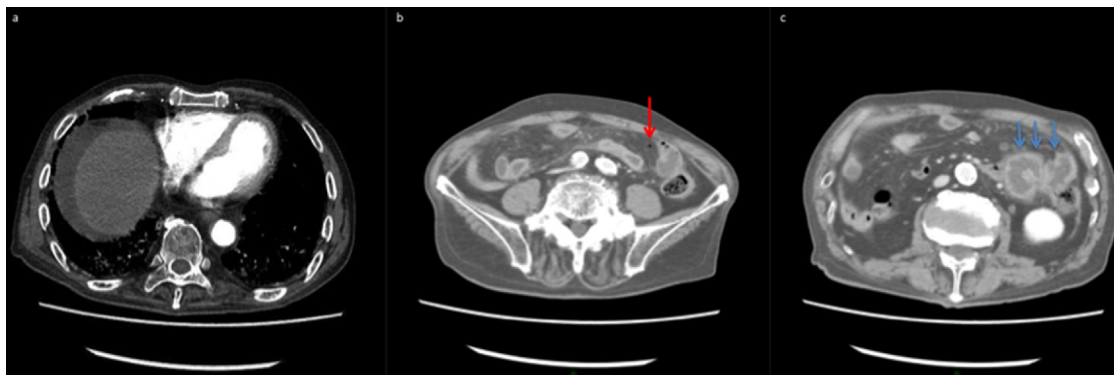


Fig. 2. A contrast CT of the abdomen showing ascites (a), free air (b; red arrow), and irregular thickness of the small intestinal walls (c; blue arrows).

(Table 1). A computed tomography (CT) scan of the chest showed marked thickening of the middle intrathoracic oesophageal wall, bilateral infiltration, and a mass, 10 × 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 80cm 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			Treatment	Other metastasis	Outcome after the metastasis (duration)
						Location	Histology	Treatment	Symptom	Number of lesions	Metastatic etiology			
1	2	1985	Wang	65	M	Lt	SCC	Res	Obstruction	1	ND	Res	None	ND
2	5	1988	Williams	60	M	Mt	SCC	None	Obstruction	1	ND	None	Lung	Dead (3 days)
3	3	1996	Yamada	56	M	Mt	SCC	Res→CRT	Obstruction	1	ND	Res	Kidney	Dead (3 years)
4	6	2005	Neve	56	M	Lt	SCC	Res→Rad	Obstruction	1	ND	Res	Chest wall	Alive (8 months)
5	8	2005	Sreenarasimhaiah	62	M	Lt	SCC	CRT	Perforation	1	ND	CRT	None	ND
6	1	2005	Lindenmann	54	M	Mt	SCC	Res→Chem	Occasionally found	1	ND	Res	None	Alive (12months)
7	10	2005	Artulraj	52	M	ND	SCC	CRT	Obstruction	1	ND	Res	Supraclavicular LN	Dead (4 months)
8	9	2009	Dasari	42	M	Lt	AC	Chem→Res	Obstruction	1	Lymph	Res	Liver	ND
9	11	2009	Horio	72	M	Lt	SCC	CRT→Res	Obstruction	1	ND	Res	Mesenteric LN	Alive (9 months)
10	12	2013	Yamada	69	M	Lt	SCC	CRT→Res	Obstruction	1	ND	Res	None	Dead (6 months)
11	4	2015	Chino	71	M	Lt	SCC	Chem→Res→Chem	Perforation	1	Lymph or Hemat	Res	None	Dead (9 months)
12	13	2017	Morinaga	72	M	Mt	SCC	Res→Chem	Obstruction	1	Lymph	Res	Lung	Alive (1 month)
13	-	2018	Our case	86	M	Mt	SCC	None	Perforation	2	Lymph or Hemat	Res	Pleural Liver Bone Lung	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.



Fig. 3. Intraoperative findings showed the perforated surface of the small intestine ulcer located 110 cm and localised ulcer 80 cm distal to the ligament of Treitz.

observed 28 cm from the incisors (Fig. 5). The pathological results of the biopsy revealed a moderately differentiated squamous cell carcinoma of the oesophagus as well (Fig. 4d). Thus, he was diagnosed with advanced oesophageal squamous cell carcinoma of the middle thoracic oesophagus. Placement of an oesophageal stent was proposed, but the patient declined any treatments and was discharged from the hospital. However, he died one month after the diagnosis due to the progression of the cancer.

3. Discussion

Metastasis of oesophageal carcinoma to the small intestine is extremely rare, and squamous cell carcinoma of the oesophagus is characterised by extensive local growth, contiguous and haematogenous spread, and lymph node involvement [2,3]. Since the first description of small intestinal metastasis from oesophageal carcinoma by Wang in 1985, some cases of small intestinal metastasis from oesophageal carcinoma have been described in the literature.

To date, 12 cases of small intestinal metastasis from oesophageal carcinoma have been reported in previous English literature [1–4,6–13]. Table 2 shows the characteristics of the patients with small intestinal metastasis from oesophageal carcinoma, including our case. All 13 patients were male, and the median age at presentation was 62 years (range, 42–86 years). Twelve cases (92%) were squamous cell carcinoma, although one case was associated with adenocarcinoma. Primary oesophageal carcinomas were located at the lower thoracic oesophagus in 7 cases (54%) but at the middle thoracic oesophagus in 5 cases (38%).

Specific symptoms of small intestinal metastasis from oesophageal carcinoma are mainly divided into obstruction and perforation. Therefore, in addition to the physical assessment, emergent morphological studies, such as CT scans, are crucial for early diagnosis. Resection of small intestinal metastasis was done in almost all patients due to these symptoms, but multiple intestinal metastases were noted in only our case.

Metastatic aetiology is still unknown. The intra-abdominal region can be connected to the oesophagus via the lymphatic network, lymphatic embolisation, and peritoneal seeding of the tumour during surgery. The haematogenous route, especially via

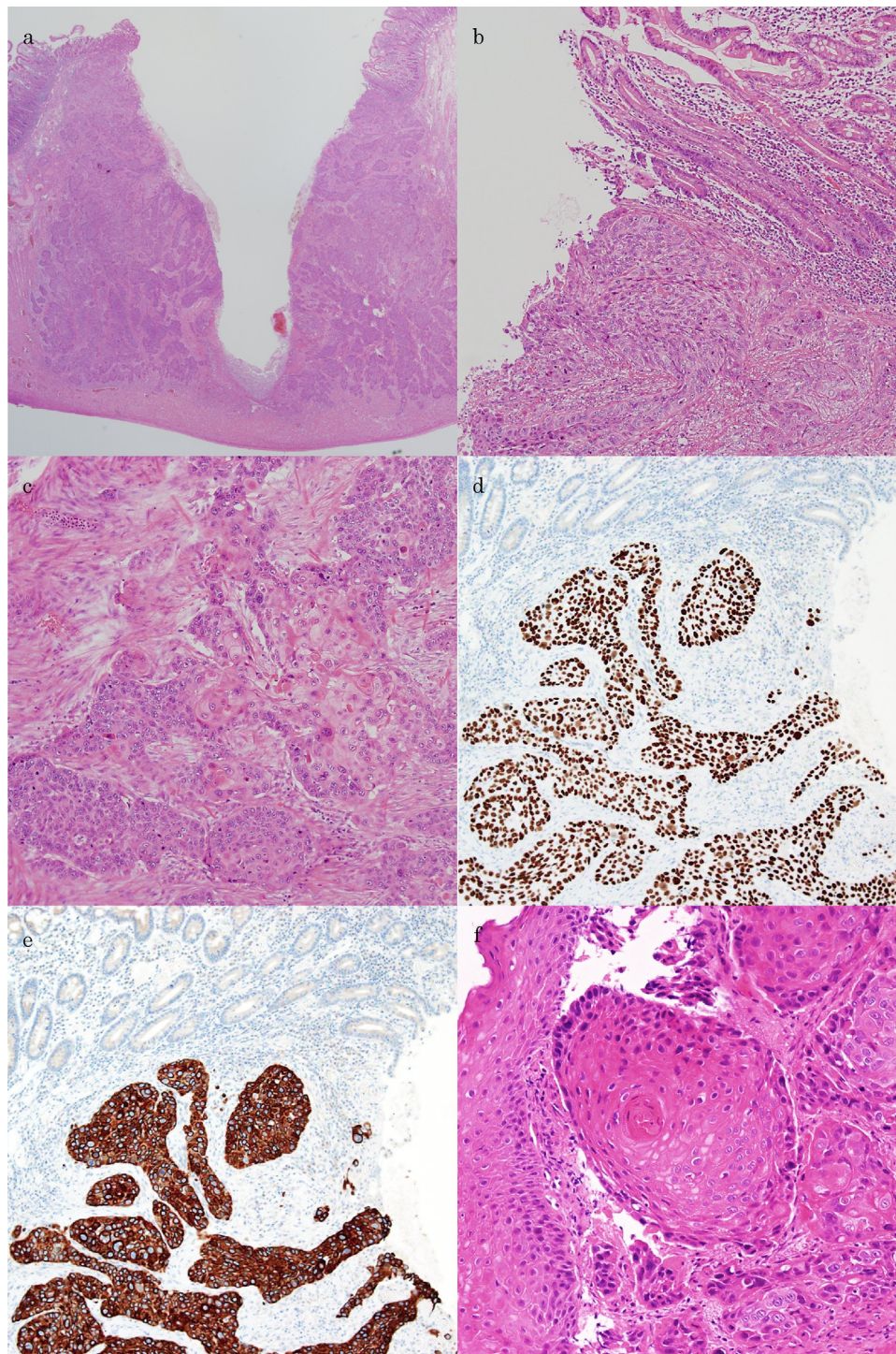


Fig. 4. The pathological findings of the resected small intestine revealing the ulcers with squamous cell carcinoma (a: Hematoxylin and eosin (HE) stain $\times 40$ magnification. b: HE stain $\times 100$ magnification. c: HE stain $\times 200$ magnification. d: P63 immunostaining $\times 100$ magnification. e: Cytokeratin 5/6 immunostaining $\times 100$ magnification.) The pathological results of the esophageal biopsy showing a moderately differentiated squamous cell carcinoma (f: HE stain $\times 200$ magnification.).

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

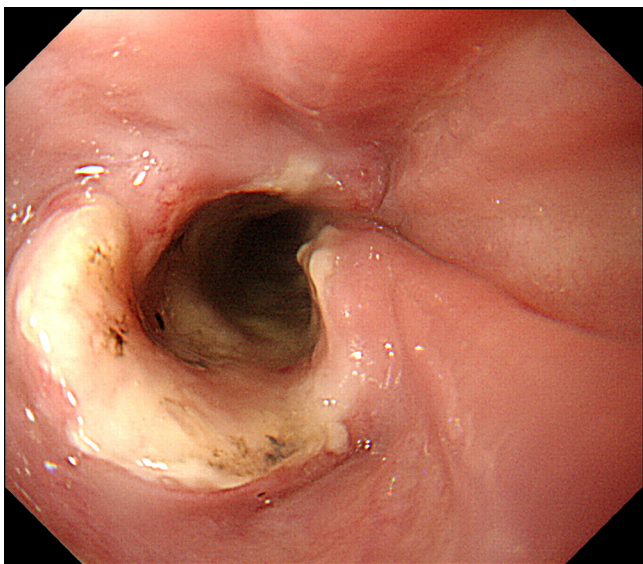


Fig. 5. An upper gastrointestinal endoscopy showing ulcerative and localised type of oesophageal tumour 28 cm from the incisors.

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.

Funding

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

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

Ryohei Ono.

References

- [1] J. Lindenmann, F. Gollwitsch, V. Matzi, C. Porubsky, A. Maier, F.M. Smolle-Juettner, Occult solitary submucosal jejunal metastasis from oesophageal carcinoma, *World J. Surg. Oncol.* 3 (2005) 44.
- [2] M. Wang, J. Patel, T.T. Casey, R. Kieffer, G.D. Dunn, Metastatic squamous cell carcinoma from the esophagus occurring as small bowel obstruction, *South. Med. J.* 78 (7) (1985) 884–886.
- [3] T. Yamada, S. Yagi, Y. Tatsuzawa, S. Fujioka, H. Sato, S. Kitagawa, et al., Small intestinal metastasis from esophageal carcinoma associated with small intestinal obstruction: report of a case, *Surg. Today* 26 (10) (1996) 800–802.
- [4] O. Chino, H. Makuuchi, S. Ozawa, H. Shimada, T. Nishi, S. Yamamoto, et al., Small intestinal metastasis from esophageal squamous cell carcinoma presenting with perforated peritonitis, *Tokai J. Exp. Clin. Med.* 40 (2) (2015) 63–68.
- [5] R.A. Agha, A.J. Fowler, A. Saeta, I. Barai, S. Rajmohan, D.P. Orgill, et al., The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [6] D.J. Williams, Metastatic oesophageal squamous carcinoma in a small bowel neurofibroma, *Jpn. J. Surg.* 18 (1) (1988) 110–113.
- [7] R.S. Neve, S.S. Qureshi, R.C. Mistry, Ileal metastases from oesophageal carcinoma causing intestinal obstruction, *J. Postgrad. Med.* 51 (1) (2005) 74–75.
- [8] J. Sreenarasimhaiah, M.P. Hoang, Esophageal squamous cell carcinoma with metastasis to the ampulla, *Gastrointest. Endosc.* 62 (2) (2005) 310–311, discussion 1.
- [9] B.V. Dasari, J. Lee, D. Reid, D. Carey, Ileocecal intussusception due to isolated metastasis from primary esophageal adenocarcinoma, *South. Med. J.* 102 (4) (2009) 419–421.
- [10] P. Arulraj, V. Damodaran, M.L. Raman, V. Nagarajan, N.R. Tulasi, Small bowel metastases from esophageal and oropharyngeal cancers, *Indian J. Gastroenterol.* 24 (3) (2005) 116–118.
- [11] T. Horio, S. Aiko, Y. Kadoma, N. Kanai, T. Ishizuka, H. Shimazaki, et al., Solitary jejunal metastasis from esophageal squamous cell carcinoma, a case report, *J. Natl. Def. Med.* 34 (1) (2009) 23–29.
- [12] I. Yamada, S. Takeno, K. Maki, Y. Sumiyoshi, T. Shiraiishi, A. Iwasaki, et al., Synchronous solitary small intestinal metastasis from esophageal squamous cell carcinoma: report of a case, *Esophagus* 10 (2013) 264–267.
- [13] T. Morinaga, K. Horio, Shimada, K. Morita, K. Yamamoto, S. Ikeshima, et al., A case report about small intestinal metastasis from esophageal carcinoma combined with bowel obstruction, *Biomed. J. Sci. Tech. Res.* 1 (6) (2017), BJSTR. MS.ID.000536.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.