CASE REPORT

Intussusception caused by small intestine metastasis of malignant pleural mesothelioma: a case report

Michinori Hamaoka1,*, Masataka Nakagawa1, Hideki Nakahara1, Rie Yamamoto2, Takashi Nishisaka2 and Toshiyuki Itamoto1

1Department of Gastroenterological Surgery, Hiroshima Prefectural Hospital, Hiroshima 734-8530, Japan and 2Department of Pathology, Hiroshima Prefectural Hospital, Hiroshima, Japan

*Correspondence address. Department of Gastroenterological Surgery, Hiroshima Prefectural Hospital, 1-5-54 Ujina-Kanda, Minami-ku, Hiroshima 734-8530, Japan. Tel: +81-82-254-1818; Fax: +81-82-253-8274; E-mail: oosajishai@gmail.com

Abstract

Malignant pleural mesothelioma (MPM) is an aggressive form of malignant tumor that originates in the pleural mesothelioma and presents as a local disease in the affected hemithorax. Small intestine metastasis is a rare complication. Herein, the case of a patient with jejunal intussusception caused by small intestine metastasis of MPM has been reported. A 72-year-old man with MPM was admitted to our hospital for abdominal pain. Computed tomography revealed small intestine intussusception. An emergency surgery was performed, and the tumor and intussusception were located in the upper jejunum. Histopathological examination of the resected jejunum revealed that the tumor was a small intestinal metastasis of the MPM from the chest wall. This case showed that MPM may metastasize to the small intestine, and metastatic tumors may cause intussusception.

INTRODUCTION

Malignant pleural mesothelioma (MPM) can invade both visceral and parietal pleura. It frequently extends to adjacent structures, such as the chest wall, mediastinum and diaphragm. Sites of lymph node spread and/or metastases include distant organs, such as the lungs, liver, kidneys, adrenal glands and brain. However, metastasis to the small intestine is rare [1]. Furthermore, intussusception caused by the metastatic MPM is a rare complication. Therefore, we present a rare case of a patient with jejunal intussusception caused by intestinal metastasis of MPM.

CASE REPORT

A 72-year-old man was diagnosed with stage IV (cT4N0M0) epithelioid MPM with diffuse invasion of soft tissues of the right chest wall, according to the criteria of International Mesothelioma Interest Group, by computed tomography (CT), positron emission tomography (PET)-CT, and pleural biopsy 18 months ago. He was treated with pemetrexed and cisplatin for 12 months. Because of the appearance of lung metastasis, the patient was treated with nivolumab as second line therapy for 6 months. He complained of vague abdominal pain 2 months ago. He then underwent upper and lower gastrointestinal endoscopy, but no abnormalities were found. He was admitted to our hospital for aggravation of abdominal pain. His abdomen was flat and soft, and he complained of abdominal pain upon applying pressure on the entire abdomen. Hematological tests revealed a raised white blood cell count of 9400/μl and high C-reactive protein level of 10.09 mg/dl. CT scan revealed intussusception of the small intestine (Fig. 1).

An emergency surgery was performed. The tumor with the intussusception was located in the upper jejunum, 120 cm distal...
to the ligament of Treitz. After the intussusception was repo-
sitioned using the Hutchinson’s maneuver, the jejunum was
resected. There was no thickening of the mesentery or dissem-
nated nodules. The postoperative course was uneventful, and
the patient was discharged on Day 9 post operation.

Histopathological examination revealed mucosal and sub-
mucosal involvement. The tumor was composed of sheets of
epithelioid cells with a high nuclear grade. On immunohisto-
chemical examination, the tumor cells tested negative for CEA,
CD34, c-kit, CK5/6 and C20, but they were positive for vimentin,
calretinin, WT-1, D2-40 and CK7 (Fig. 2). The results were identi-
cal to those of the MPM from the chest wall. Thus, we concluded
that the tumor was a metastasis of MPM.

DISCUSSION

MPM is an aggressive form of malignant tumor that originates
in the pleural mesothelioma. MPM typically invades surround-
ning organs and rarely metastasizes. However, upon reviewing
autopsy cases, extrathoracic metastasis was found in 55% of
the cases. The reported metastatic sites included almost all
organs, such as the lymph nodes, lungs, adrenal glands, liver,
kidneys and brain [1]. Metastases to the small intestine are
infrequent. Upon reviewing published literature, seven cases of
small intestine metastasis from MPM were found (Table 1)[2–
8]. Diagnosing this condition before symptoms occur is difficult.
In many cases, the diagnosis was made after the appearance of
symptoms, such as anemia or perforation. Gocho et al. [4] have
suggested the following reasons why detecting small intestine
metastases is difficult. First, physicians have little knowledge of
clinically rare metastases, such as small intestine metastases.
Second, non-specific symptoms may be considered as general
complaints or side effects of chemotherapy. Finally, follow-up
CT scans have low sensitivity for small intestine tumors [4].
Similarly, the patient in our case complained of vague abdomi-
nal pain for ∼2 months, and the small intestine metastasis could
not be diagnosed before detecting the intussusception. Navaro
et al. [7] have suggested that PET-CT as well as the combina-
tion of capsule endoscopy and double-balloon enteroscopy may aid
in overcoming difficulties in detecting this type of metastasis. If
patients with mesothelioma have unexplained abdominal pain,
these tests should be considered for the detecting small intestine
metastases.

According to Marsicovetere et al. [9], intussusception accounts
for only 1–5% of ileus cases. Adult cases of intussusception are
rarer than pediatric cases as they account for 5–10% of all intus-
susceptions. In total, 90% of the symptomatic cases have been
associated with identifiable causes, whereas the remaining 10%
Intussusception caused by small intestine metastasis of MPM

have been declared as idiopathic cases. Most intussusception in adults arises from the small intestine, and 50-75% of lesions are benign. Malignant intraluminal causes of small intestine intussusception include primary leiomyosarcomas, adenocarcinoma, gastrointestinal stromal tumors, carcinoid tumors, neuroendocrine tumors and lymphomas [9]. Less commonly, small intestine metastasis may act as lead points of the intussusception. Although there were several reports of small intestine intussusception due to metastatic disease such as melanoma, lung cancer and renal cell cancer, MPM was extremely rare.

The main routes of metastasis to the small intestine include direct invasion, intraperitoneal seeding and hematogenous metastases. In this case, since there was no evidence of peritoneal dissemination, metastasis may have occurred through the hematogenous route. Distant hematogenous metastases usually appear during the late disease stages [10]. In half of the cases, the period from the diagnosis of MPM to the appearance of symptoms due to small intestine metastases has passed ~1 year (Table 1). Histologically, MPM is classified as epithelioid (60%), biphasic (30%) or sarcomatoid (10%). Sarcomatoid mesothelioma is associated with more frequent distant metastases than the others. Although this case was an epithelioid type MPM with a relatively good prognosis, distant metastasis might have occurred, with a long-term survival of a year and a half.

Figure 2: A tumor of size 35 mm is found from the mucosal to the subserosal layer of the jejunum (a, b, and c); the tumor is composed of sheets of epithelioid cells with high nuclear grade (×20 magnification hematoxylin and eosin staining) (d); immunohistochemical examination reveals tumor cells positive for vimentin (e), calretinin (f), WT-1 (g), D2-40 (h) and CK7 (i), and negative for CEA (j), CD34 (k), c-kit (l), CK5/6 (m) and CK20 (n).
In conclusion, MPM is a disease that can cause metastasis to the small intestine. Therefore, if a patient with MPM develops abdominal pain, metastasis to the small intestine should be considered.

CONFLICT OF INTEREST STATEMENT
None declared.

FUNDING
None.

REFERENCES
1. Finn RS, Brims FJH, Gandhi A, Olsen N, Musk AW, Maskell NA, et al. Postmortem findings of malignant pleural mesothelioma: a two-center study of 318 patients. Chest 2012;142:1267–73.
2. Kakugawa Y, Watanabe S, Kobayashi N, Tani M, Tanaka S, Tsuta K, et al. Diagnosis of small-bowel metastasis of malignant pleural mesothelioma by capsule endoscopy and double balloon enteroscopy. Endoscopy 2007;39:E229–30.
3. Chen HC, Tsai KB, Wang CS, Hsieh TJ, Hsu JS. Duodenal metastasis of malignant pleural mesothelioma. J Formosan Med Assoc 2008;107:961–4.
4. Gocho K, Isobe K, Kaburaki K, Honda Y, Mitsuda A, Akasaka Y, et al. Malignant pleural mesothelioma presenting as an acute surgical abdomen due to metastatic jejunal perforation. Internal Med (Tokyo, Japan) 2010;49:597–601.
5. Martínez Caselles A, Baños Madrid R, Egea Valenzuela J, Molina Martínez J, Carballo ÁF. Gastrointestinal bleeding secondary to duodenal metastases of malignant pleural mesothelioma. Revista Espanola Enfermedades Digestivas 2010;102:602–3.
6. Liu H, Cheng YJ, Chen HP, Hwang JC, Chang PC. Multiple bowel intussusceptions from metastatic localized malignant pleural mesothelioma: a case report. World J Gastroenterol: WJG 2010;16:3984–6.
7. Navarro García MI, Sánchez Pérez A, Vázquez Rojas JL. Jejunal perforation by metastasis of malignant pleural mesothelioma. Arch Bronconeumol 2015;51:366–7.
8. Alkhayal K. Metastatic malignant pleural mesothelioma masquerading as a case of acute abdomen secondary to small bowel perforation. Ann Saudi Med 2016;36:229–31.
9. Marsicovetere P, Ivatury SJ, White B, Holubar SD. Intestinal intussusception: Etiology, diagnosis, and treatment. Clin Colon Rectal Surg 2017;30:30–9.
10. Brenner J, Sordillo PP, Magill GB, Golbey RB. Malignant mesothelioma of the pleura: review of 123 patients. Cancer 1982;49:2431–5.