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Case Report

Recurrent Intussusception due to Multiple Intestinal Metastases From Lung Adenocarcinoma

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Metastasis to the small intestine presents occasionally as multiple lesions and is associated with numerous complications. However, metastasis-induced intussusception in adults is a rare phenomenon. We report a case of recurrent intussusception induced by metastatic lesions from lung cancer. A 54-year-old male was referred to our hospital with intermittent lower right abdominal pain. Chest computed tomography (CT) revealed a mass, suggesting lung cancer, and abdominal CT showed characteristic target-shaped lesions highly suggestive of enteric intussusception in the ileum. The patient underwent segmental resection of the intussuscepted ileum, and analysis of the resected specimen identified a poorly differentiated adenocarcinoma that was pathologically and immunohistologically identical to the lung specimen obtained by percutaneous needle biopsy. Although the symptoms transiently resolved after surgery, intussusception recurred 3 weeks later, and the patient died 28 days after surgery. Multiple metastatic lesions should be considered in adult intussusception, particularly in patients with a history of malignancy.

Key words: Intussusception – Intestinal metastasis – Lung adenocarcinoma

Intussusception occurs primarily in pediatric patients and is the second most common cause of abdominal emergencies.[1] In contrast, adult intussusception is rare, constituting 5% of all intussusception cases; it is the underlying etiology in only 1% of all adult bowel obstruction cases.[2] The majority of cases in adults are due to definable structural lesions[3–5]; however, most are benign, and

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malignancy-mediated intussusception is rare.[6] On the other hand, lung cancer is one of the most frequently diagnosed cancers and is the leading cause of malignancy-related deaths worldwide. Metastases to the liver, adrenal gland, central nervous system, and bone are common in patients with lung cancer, whereas metastasis to the small intestine is rare.[7] Here we report a case of a patient presenting with recurrent intussusception induced by multiple metastatic lesions from lung adenocarcinoma.

Case Report

A 54-year-old male was referred to the emergency department with a 2-week history of intermittent lower right abdominal pain accompanied by nausea and vomiting. The patient was an active smoker with 40 cigarettes per day for 30 years. He had bloody sputum and cough more than 6 months ago. On admission, his laboratory results, including carcinoembryonic antigen (CEA), were within the normal limits except for white blood cell count (9400/μL), C-reactive protein (9.0 mg/dL), albumin (3.0 g/dL), blood urea nitrogen (BUN, 28.4 mg/dL), serum Na (131 mEq/L), serum Cl (88 mEq/L), and carbohydrate antigen 19-9 (CA19-9, 383.7 U/mL). His blood gas levels were poor, with arterial oxygen saturation of 91.1% and pO_2 of 56.3 mmHg. Chest computed tomography (CT) revealed a mass, suggesting lung cancer with thoracic vertebral invasion, hilar lymph node metastases, and atelectasis of the lower lobe extending from the hilar region of the right lung (Fig. 1a). Abdominal ultrasonography revealed increased intestinal wall thickness with a target sign in the right lower quadrant area (Fig. 1b). Abdominal CT scan was performed to confirm the ultrasonography findings: a three-layered structure with the characteristic target-shaped appearance highly suggestive of enteric intussusception in the ileum (Fig. 1c). Furthermore, CT indicated metastatic lesions in the right adrenal gland, pancreas, bilateral kidneys, and para-aortic lymph nodes. The patient was severely dehydrated due to continuous vomiting over the previous weeks. Decompression with an ileus tube and correction of biochemical abnormalities with intravenous resuscitation were performed as initial management. After improvement of his overall status was confirmed, laparotomy was performed under spinal anesthesia due to poor respiratory function, which led to the diagnosis of intussusception. The patient underwent resection of the ileal segment showing intussusception (Fig. 2), and an ileostomy with double orifices was created without anastomosis. Macroscopic findings of the resected specimen included a tumor on the ileal wall measuring 2.5 × 2.0 × 2.0 cm that occupied more than one-third of the circumference. Histologic analysis identified a poorly differentiated adenocarcinoma proliferating mainly in the submucosa and muscular layer and invading the subserosa with the absence of regional lymph node metastasis (Fig. 3a). Moderate lymphangitic and venous invasion were observed according to the Japanese classification of colorectal carcinoma.[8] Carcinoma cells were diffusely positive for cytokeratin (Fig. 3b), vimentin, thyroid transcription factor 1 (Fig. 3c), and napsin A by immunohistochemistry. The resected ileum and the lung specimen (Fig. 3d) obtained by percutaneous needle biopsy were pathologically identical. Based on these findings, the patient was diagnosed with primary lung adenocarcinoma with metastasis to the ileum. Following surgical treatment, the symptoms resolved transiently. Radiotherapy to the mediastinal lesion was started 9 days after surgery. However, the tumor mass increased rapidly, and pleural effusion developed. Furthermore, intussusception recurred 3 weeks after the surgery. The patient did not receive any further treatment except for nasogastric drainage by a Ryles tube and pain control; he died 28 days after surgery.

Discussion

Adult intussusception was first reported in 1674 by Barbette[9] and further presented in a detailed report in 1789 by John Hunter as "intussusception."[10] Intussusception is defined as the telescoping of a proximal segment of the gastrointestinal tract into the lumen of the adjacent distal segment of the gastrointestinal tract.[3] Although the exact mechanism of intussusception is unknown, any lesion in the bowel wall or irritant within the lumen that alters normal peristaltic activity is suggested to initiate an invagination. Intussusception in children is usually primary; there are no pathologic lead points in the majority of children with intussusception.[3] However, approximately 90% of adult cases of intussusception are secondary to a pathologic condition that serves as a lead point.[3–5] The majority of identifiable causes of small bowel intussusceptions are from benign lesions, such as hamartomatous polyps, Meckel's diverticula, and strictures. Malignancy is found in 6% to 30% of cases of small bowel intussusceptions in adults.[6] These malignan-

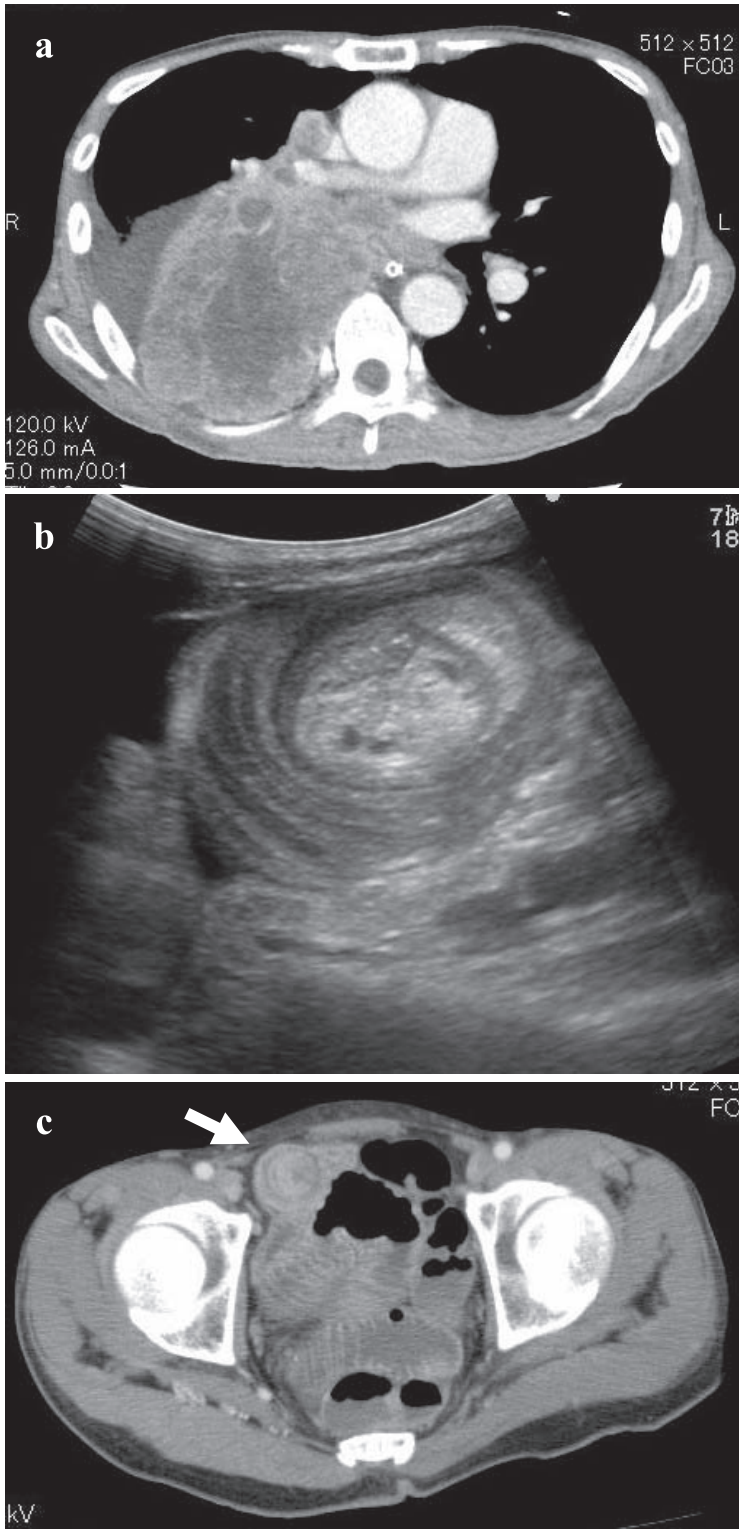

Fig. 1 (a) Chest computed tomography (CT) showed a mass suggesting lung cancer with thoracic vertebral invasion, hilar lymph node metastases, and atelectasis of the lower lobe extending from the hilar region of the right lung. (b) Abdominal ultrasonography showed increased intestinal wall thickness with a target sign in the right lower quadrant. (c) Abdominal CT showed a 3-layered structure with the characteristic target-shaped appearance, highly suggestive of enteric intussusception in the ileum.

cies include primary lesions, such as lymphoma or gastrointestinal tumor, and metastases, in particular from the lung, breast, and malignant melanoma.[6,11] Yang *et al* reported symptomatic gastrointestinal metastasis in 1% of patients with primary lung cancer.[12] On the other hand, McNeil *et al* found that 10% of lung cancer patients had small bowel metastases at postmortem, all of which were

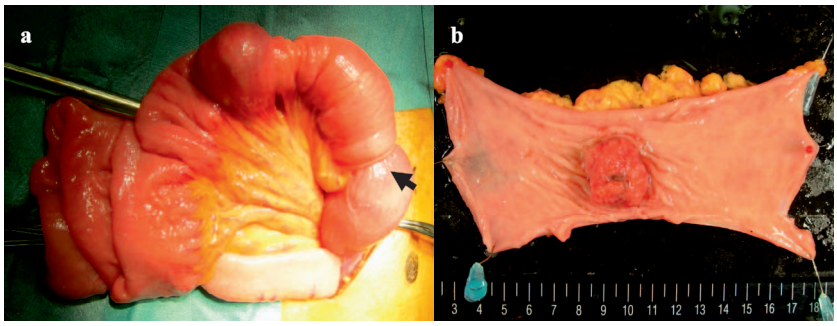

Fig. 2 (a) Intraoperative findings of small bowel intussusception (b) Macroscopic findings of the resected specimen showed a tumor in the ileal wall measuring 2.5 × 2.0 × 2.0 cm and occupying more than one-third of the circumference.

asymptomatic.[13] Metastasis from the lungs to the gastrointestinal tract is reportedly more common in large cell carcinoma than in other types of cancer;[7] however, the intestinal clinical features do not differ by the histologic type. Intestinal metastasis from primary lung cancer is a sign of widespread metastatic disease and poor prognosis.[14] Our patient had metastatic lesions in multiple organs, his condition worsened rapidly, and he died 38 days after admission to our hospital.

Ultrasonography is considered a useful tool for the diagnosis of intussusception, although obesity and presence of massive air in the distended bowel loop can limit image quality and subsequent diagnostic accuracy.[15] CT is currently considered as the most sensitive radiologic method to confirm intussusception.[2,16] In addition, CT can define the location and nature of the mass and its relationship with the surrounding tissues; it may also help in the staging of patients with suspected malignancy causing the intussusception. In the present case, ultrasonography and CT showed the characteristic signs of intussusception and were useful in diagnosis. Furthermore, CT was effective in diagnosing the primary malignant lesion causing the intussusception. Adult intussusceptions are managed mainly through surgery due to the high probability of an organic lead point. Several case reports have demonstrated that multiple lesions may cause recurrent intussusceptions, particularly in malignancies such as lymphoma, melanoma, and metastases from other sites.[14,17,18] In the present study, a second intussusception due to a separate metastatic lesion originating from the lung cancer occurred early after surgery, although no other lesions were found within the intestine during surgery. Inspection of the intestine during surgery was challenging due to spinal anesthesia; however, in adult cases of intussusception, thorough abdominal inspection

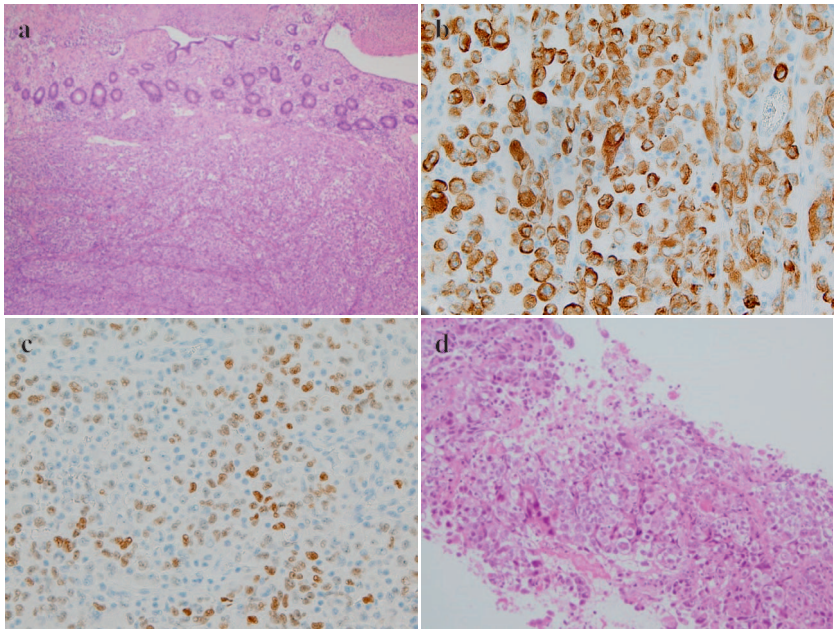

Fig. 3 (a) Histologic analysis identified a poorly differentiated adenocarcinoma proliferating mainly in the submucosa and muscular layer and invading the subserosa (hematoxylin and eosin staining, ×40 magnification). (b) Immunohistochemical staining of the cancerous cells showed diffuse positivity for cytokeratin (×200 magnification). (c) Immunohistochemical staining of the cancerous cells showed diffuse positivity for thyroid transcription factor 1 (×200 magnification). (d) The resected ileum and the lung specimen obtained by percutaneous needle biopsy were pathologically identical (hematoxylin and eosin staining, × 40 magnification).

may be necessary to rule out multiple lesions that can later lead to recurrence.

In conclusion, malignant metastatic lesions should be taken into consideration as a cause of adult intussusception. Furthermore, it is critical to consider multiple metastatic lesions as a cause of potential recurrence, particularly in patients with a history of malignancy.

Acknowledgments

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References

1. Lin MW, Wu CT, Chang YL. Intussusception caused by intestinal metastasis from lung pleomorphic carcinoma. *Ann Thorac Cardiovasc Surg* 2014;**20 Suppl**:635–638
2. Azar T, Berger DL. Adult intussusception. *Ann Surg* 1997;**226**(2):134–138
3. Marinis A, Yiallourou A, Samanides L, Dafnios N, Anastasopoulos G, Vassiliou I *et al*. Intussusception of the bowel in adults: a review. *World J Gastroenterol* 2009;**15**(4):407–411
4. Stubenbord WT, Thorbjarnarson B. Intussusception in adults. *Ann Surg* 1970;**172**(2):306–310
5. Weilbaecher D, Bolin JA, Hearn D, Ogden W, 2nd. Intussusception in adults. Review of 160 cases. *Am J Surg* 1971;**121**(5):531–535
6. Begos DG, Sandor A, Modlin IM. The diagnosis and management of adult intussusception. *Am J Surg* 1997;**173**(2):88–94
7. Rossi G, Marchioni A, Romagnani E, Bertolini F, Longo L, Cavazza A *et al*. Primary lung cancer presenting with gastrointestinal tract involvement: clinicopathologic and immunohistochemical features in a series of 18 consecutive cases. *J Thorac Oncol* 2007;**2**(2):115–120
8. Japan Society for Cancer of the Colon and Rectum, eds. *Japanese classification of colorectal carcinoma*. 2nd English ed. Tokyo, Japan: Kanehara & Co, 2009
9. de Moulin D. Paul Barbette, M.D.: a seventeenth-century Amsterdam author of best-selling textbooks. *Bull Hist Med* 1985;**59**(4):506–514
10. Noble I, ed. Master surgeon: John Hunter. New York: J Messner, 1971
11. Wang LT, Wu CC, Yu JC, Hsiao CW, Hsu CC, Jao SW. Clinical entity and treatment strategies for adult intussusceptions: 20 years' experience. *Dis Colon Rectum* 2007;**50**(11):1941–1949
12. Yang CJ, Hwang JJ, Kang WY, Chong IW, Wang TH, Sheu CC *et al*. Gastro-intestinal metastasis of primary lung carcinoma: clinical presentations and outcome. *Lung Cancer* 2006;**54**(3):319–323
13. McNeill PM, Wagman LD, Neifeld JP. Small bowel metastases from primary carcinoma of the lung. *Cancer* 1987;**59**(8):1486–1489
14. Morris DM, Deitch EA. Clinically significant intestinal metastasis from a primary bronchogenic carcinoma. *J Surg Oncol* 1983;**23**(2):93–94
15. Fujii Y, Taniguchi N, Itoh K. Intussusception induced by villous tumor of the colon: sonographic findings. *J Clin Ultrasound* 2002;**30**(1):48–51
16. Gayer G, Apter S, Hofmann C, Nass S, Amitai M, Zissin R *et al*. Intussusception in adults: CT diagnosis. *Clin Radiol* 1998;**53**(1):53–57
17. Bak M, Teglbjaerg PS. Pleomorphic (giant cell) carcinoma of the intestine. An immunohistochemical and electron microscopic study. *Cancer* 1989;**64**(12):2557–2564
18. Kouladouros K, Gartner D, Munch S, Paul M, Schon MR. Recurrent intussusception as initial manifestation of primary intestinal melanoma: Case report and literature review. *World J Gastroenterol* 2015;**21**(10):3114–3120