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CASE REPORT

Jejunal intussusception caused by metastasis of a giant cell carcinoma of the lung

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SUMMARY

A 55-year-old woman was admitted to our hospital reporting of nausea, vomiting and anorexia. One month before admission, she had been diagnosed with lung cancer with intestinal metastasis. A CT scan confirmed intussusception due to intestinal metastasis and she underwent emergency laparoscopic surgery followed by resection of the primary lung cancer. Histopathological findings of the intestinal specimen suggested the metastasis was from a giant cell carcinoma of the lung, which had extensive necrosis. She was still alive without recurrence 11 months after the first surgery. Giant cell carcinoma of the lung is a rare type of non-small cell carcinoma and intestinal metastasis is one of the unique features. This type of tumour has such aggressive characteristics that oncological prognosis is reported to be extremely poor. In our case, however, complete surgical resection of both primary and metastatic tumours might result in a better outcome than has been reported.

BACKGROUND

Giant cell carcinoma of the lung is a rare form of poorly differentiated non-small cell lung carcinoma (NSCLC), which is classified as a sarcomatoid carcinoma according to the 2015 WHO classification.[1] Giant cell carcinomas are composed entirely of giant cells and have specific patterns that do not resemble those of adenocarcinomas, squamous cell carcinomas or large cell carcinomas.[1] These types are rare and account for ~0.1–0.4% of all lung carcinomas.[2–5] They are aggressive tumours and are prone to metastasize to other organs, including unusual locations such as the gastrointestinal tract and the retroperitoneal space. However, intussusception caused by the metastasis of a giant cell carcinoma of the lung is an extremely rare complication. Here, we present a rare case of a patient with jejunal intussusception caused by intestinal metastasis of a giant cell carcinoma of the lung.

CASE PRESENTATION

A 55-year-old woman who had monoclonal gammopathy of undetermined significance underwent screening using a whole-body CT scan and positron emission tomography (PET)-CT to rule out malignant myeloma. The whole-body CT scan showed a 36 mm sized mass on the left upper lung lobe and a mass forming wall thickness in the upper jejunum with an 8 mm lymph node swelling near the intestinal mass (figure 1A, B). A protruded tumour was detected in the upper jejunum using small bowel endoscopy (figure 2), and PET-CT revealed abnormal accumulation at the same lesion (figure 3). The fine-needle aspiration specimen showed that the tumour cells had large abundant cytoplasm that was densely eosinophilic, reminiscent of an epithelioid rhabdomyosarcoma, malignant melanoma or poorly differentiated carcinoma. The laboratory test revealed a highly elevated white cell count count (38 700/mm^3).

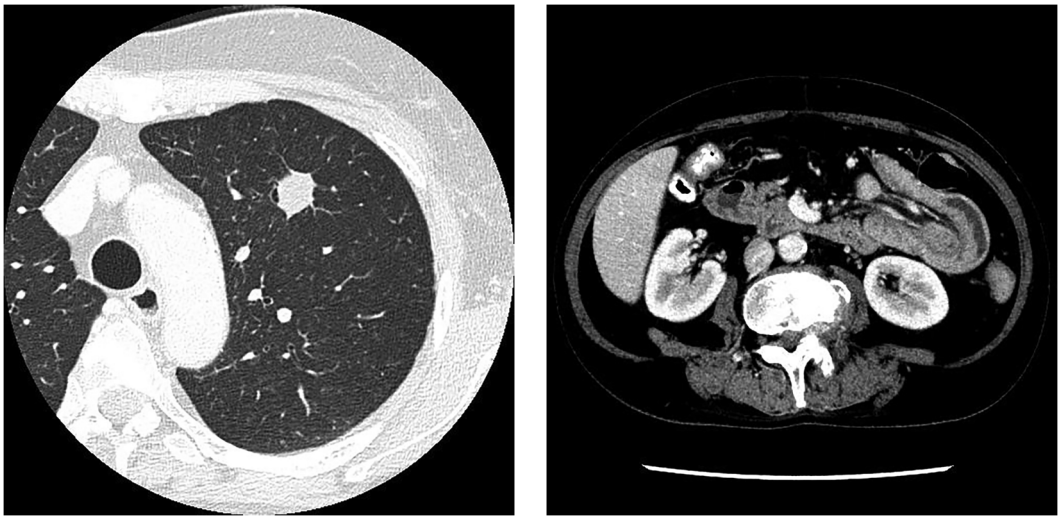

Figure 1 (A) Chest CT scan showed a 36 mm mass in the left upper lobe of the lung. (B) An abdominal CT scan showed a mass-forming wall thickness in the upper jejunum and a mesenteric lymph node swelling.

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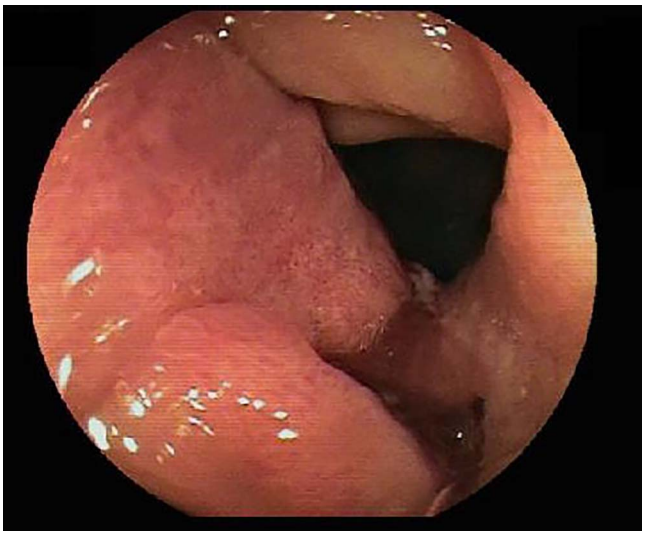

Figure 2 A small-bowel endoscopy revealed a protruded lesion in the upper jejunum.

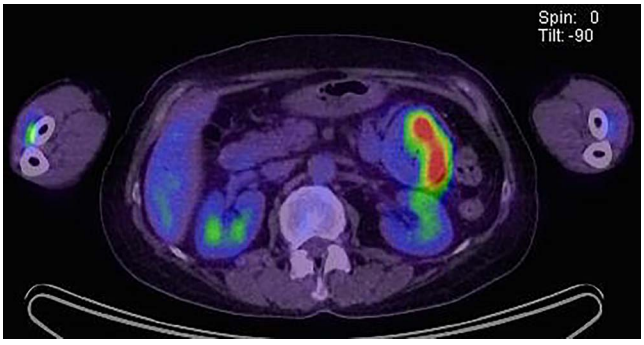

Figure 3 ^{18}F-fluorodeoxyglucose positron emission tomography revealed abnormal accumulation in the upper jejunum.

DIFFERENTIAL DIAGNOSIS

Considering the histological findings of the tumour and a lung lesion on CT scan, jejunal metastasis of the granulocyte-colony stimulating factor (G-CSF) secreting giant cell carcinoma of the lung was suspected. Other differential diagnoses were jejunal adenocarcinoma, malignant melanoma and anaplastic large cell lymphoma.

TREATMENT

One month later, she was admitted to our hospital reporting of nausea, vomiting and anorexia. We repeated the CT scan and diagnosed her with an intussusception of the jejunal tumour. We performed emergency laparoscopic surgery. The tumour with the intussusception was located in the upper jejunum 30 cm distal to the ligament of Treitz. After the intussusception was repositioned using the Hutchinson manoeuver, the jejunum was resected. The postoperative course was uneventful and the white cell count count at day 3 was decreased significantly to baseline after resection. Two months later, she underwent segmentectomy of the left upper lobe of the lung.

Histopathologically, the jejunal tumour was composed of a diffuse proliferation of tumour cells without a clear direction of differentiation, and relatively non-cohesive, pleomorphic mononucleated cells admixed with bizarre, frequently multinucleated giant cells that contained abundant eosinophilic cytoplasm. In addition, an increased number of tumour-infiltrating neutrophils and focal tumour cell emperipolesis were present (figure 4A, B). On immunohistochemical study, most tumour cells expressed cytokeratin (AE1/AE3 and CAM 5.2), vimentin and thyroid transcription factor-1 (TTF-1), while some expressed Napsin A and G-CSF. Although the immunopositivity of TTF-1 and Napsin A was supportive of metastasis from lung, the microscopic finding of the lung tumour was extensive necrosis with only scanty degenerated tumour cells remaining (figure 5). These histological findings corresponded with jejunal metastasis of a G-CSF-producing giant cell carcinoma of the lung.

OUTCOME AND FOLLOW-UP

The patient has been recurrence-free for 11 months since the first surgery. Although we planned to treat with adjuvant chemotherapy, which was the same as that used for treating NSCLC, she refused it and instead received only a follow-up examination.

DISCUSSION

Sarcomatoid carcinomas are a group of poorly differentiated NSCLCs that contain a component of sarcoma or sarcoma-like (spindle and/or giant cell) differentiation. Pleomorphic carcinomas, spindle cell carcinomas, giant cell carcinomas, carcinosarcomas and pulmonary blastomas are classified into this group according to the recent WHO classification.[1] Sarcomatoid carcinomas are very rare diseases and have unique clinical features compared with other non-small cell carcinomas. As seen in our

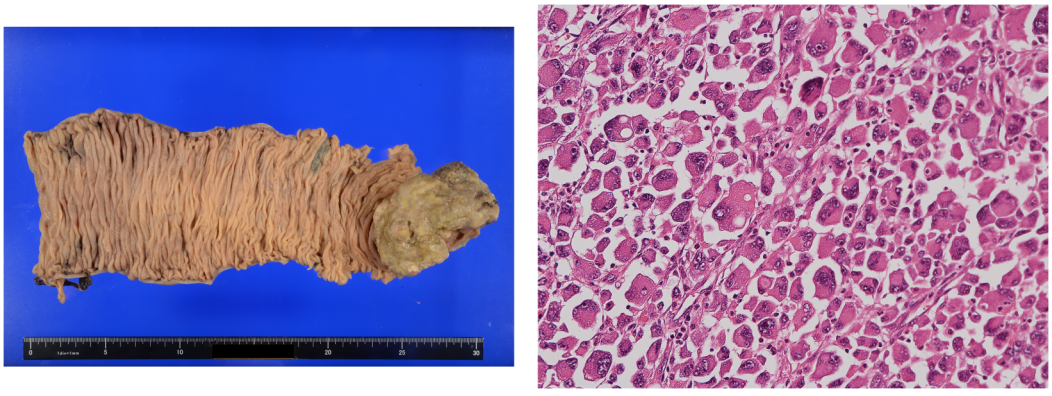

Figure 4 (A) The resected specimen of the jejunum. A protruded 89 mm mass can be seen to be invaginated into the distal jejunum. (B) The jejunal tumour was composed of a diffuse proliferation of tumour cells without a clear direction of differentiation, and relatively non-cohesive, pleomorphic mononucleated cells admixed with bizarre, frequently multinucleated giant cells that contained abundant eosinophilic cytoplasm. In addition, an increased number of tumour-infiltrating neutrophils and focal tumour cell emperipolesis were present.

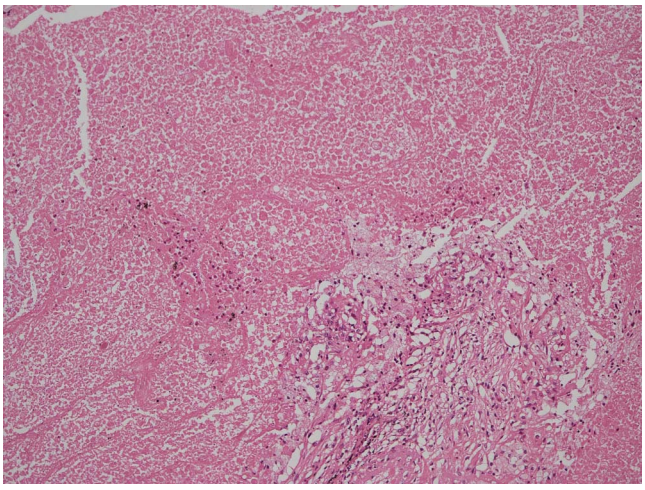

Figure 5 The microscopic findings of the lung tumour showing extensive necrosis with only scanty degenerated tumour cells remaining.

case, sarcomatoid carcinomas sometimes cause an elevated inflammatory response due to the production of G-CSF and interleukin 6.[6] Immunohistologically, TTF-1 and cytokeratin are detected in a significant percentage of sarcomatoid carcinomas.[2] The outcome is significantly poorer than that of patients with other NSCLCs, and adjuvant chemotherapy and radiotherapy seem to be ineffective. Their aggressive characteristics such as large tumours, advanced-stage cancer and pleural invasions, result in higher rates of recurrence and lower survival rates.[3 5] Some previous reports showed that the median survival time of patients with sarcomatoid carcinoma was 8–22.8 months and the 5-year survival rate was 10–36.7%.[2–5 7]

Giant cell carcinomas of the lung sometimes metastasize to the gastrointestinal tract. Wellmann *et al*[8] demonstrated that 17–25% of patients with pulmonary giant cell carcinomas had gastrointestinal metastases. However, intussusception caused by the intestinal metastasis of the primary lung carcinoma is rarely seen. Intussusception in adults accounts for <5% of all intussusceptions and in 90% of adult cases is secondary to bowel

Table 1 25 reported cases of secondary intussusceptions caused by primary lung malignancies.

Author	Published year	Age	Sex	Cancer cell type	Tumour stage at initial diagnosis	Other metastasis sites	Time interval from initial diagnosis to intussusception	Time interval from intussusception to death
Katz *et al*[16]	1981	68	M	Giant cell carcinoma	4	Liver, supraclavicular lymph node	1.6 months	NS
Listrom *et al*[17]	1988	63	M	Squamous cell carcinoma, spindle cell type	4	Left adrenal gland	Simultaneously	NS
Eng and Sabanathan[18]	1992	60	M	Carcinosarcoma	2B	Brain	18 months	6 months
Issa and Mullen[19]	1992	61	M	Adenocarcinoma, large cell type	4	Left 4th rib	1 month	1 month
Testini *et al*[20]	2002	44	M	Malignant melanoma	4	Brain	Simultaneously	12 months (still alive)
Yang *et al*[21]	2006	57	M	Small cell carcinoma	4	Brain, ascite	2 months	3 months
Alvarez *et al*[22]	2006	50	M	Big cell carcinoma	4	Vertebra	NS (but metachronous)	NS
Goh *et al*[23]	2007	57	M	Pleomorphic associated with adenocarcinoma	NS	Para-aortic lymph node, liver	3 months	7 months
		40	M	Large cell carcinoma	NS	Brain, bone	4 months	5 months
		69	M	Large cell carcinoma	NS	Bone, liver, cervical lymph node	6 months	9 months
Kagohashi *et al*[9]	2007	59	M	Large cell carcinoma	4	None	Simultaneously	6 months
Pollheimer *et al*[13]	2009	65	F	Pleomorphic carcinoma	4	NS	Simultaneously	NS
Chiu *et al*[24]	2009	87	M	Poorly differentiated adenocarcinoma	NS	Gingiva	Simultaneously	2.7 months
Shi *et al*[14]	2009	61	M	Pleomorphic sarcomatoid carcinoma	4	Right adrenal gland, liver, brain	17 months	4 months
Kini *et al*[25]	2010	78	M	Adenocarcinoma	NS	None	7 months	5 months (still alive)
Otera *et al*[10]	2010	63	M	Large cell carcinoma	4	None	Simultaneously	0.9 months
Lee *et al*[26]	2010	74	M	Poorly differentiated adenocarcinoma	4	None	Simultaneously	NS
Rashid *et al*[27]	2011	57	M	Pleomorphic carcinoma	4	None	Simultaneously	3 months
Jarmin *et al*[11]	2012	75	M	Small cell carcinoma	4	None	Simultaneously	NS
Guner *et al*[28]	2012	71	M	Sarcomatoid carcinoma	3A	None	12 months	9 months
Wou *et al*[15]	2013	77	F	Non-small cell carcinoma	4	NS	Almost simultaneously	2 months
Lin *et al*[29]	2014	78	M	Pleomorphic carcinoma	4	None	Simultaneously	3 months
Jung *et al*[30]	2014	63	M	Pleomorphic carcinoma	3A	Bone, mediastinal lymph node	5 months	NS
Mandeville *et al*[12]	2015	49	F	Giant cell carcinoma	4	Vertebra, cervical lymph node, brain	Simultaneously	6 months
Present case	2015	55	F	Giant cell carcinoma	4	None	1 month	11 months (still alive)
								NS

NS, not stated.

diseases.[9–12] Malignancy is found in 6–30% of small bowel intussusceptions following surgical resection, and lung cancers account for 21.6% of these cases.[13–15]

We examined the prevalence of secondary intussusceptions caused by primary lung malignancies by performing a PubMed search up until November 2015 using the key words 'intussusception' and 'lung cancer/carcinoma'. Inclusion criteria were case reports, case series and cohort studies that investigated patients with intussusception caused by primary lung malignancies. Exclusion criteria included studies in languages other than English. We found 25 cases, including our case, which corresponded to the criteria presented in 22 publications.[9–30] The data are shown in table 1. The male to female ratio was 21:4 and the average age was 63.2 years old (40–87 years old). The most common tumour types of the primary lung lesion were sarcomatoid carcinomas (44%), including giant cell carcinomas (12%), followed by large cell carcinomas (20%), adenocarcinomas (16%), small cell carcinomas (8%), squamous cell carcinomas (4%), non-small cell carcinomas (4%) and malignant melanomas (4%). This result supports the fact that sarcomatoid carcinomas are more likely to develop intestinal metastases. Almost all cases were diagnosed in advanced stages and intussusception was the first symptom in 44% (11/25) of all cases. The prognosis of patients with intussusception was particularly poor because of metastatic lung cancer. The median survival time after diagnosis of intussusception was 5 months (range, 0.9–12 months).

Accordingly, we considered that the intussusception because of lung cancer metastases was particularly rare, and prognoses of such cases were extremely poor. In contrast, our patient was still alive without recurrence for 11 months after the first surgery. Although the exact reason remains unclear, it seems possible that the better prognosis of this patient was due to complete resection of both primary lung lesion and intestinal metastasis. Shoji *et al*[31] reported a case of a pulmonary giant cell carcinoma with metastases to the small intestine, which resulted in more than 3-year disease-free survival after aggressive surgical resection and chemotherapy. Although the standard therapy for giant cell carcinomas of the lung with intestinal metastasis has not been established, we believe that aggressive surgical resection is an effective strategy and plays an important role in the management of such cases.

Learning points

- Giant cell carcinomas of the lung are rare tumours whose oncological prognoses are extremely poor and intestinal metastasis is one of the unique features of them.
- Intussusception because of intestinal metastasis of lung cancer is a rare complication.
- Although pulmonary giant cell carcinomas with intestinal metastases have a poor prognosis, complete surgical resection of tumours might result in a better outcome.

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REFERENCES

1 Travis WD, Brambilla E, Burke AP, *et al*. *World health organization classification of tumours of the lung, pleura, thymus and heart*. 4th edn. Lyon, France: IARC Press, 2015.
2 Rossi G, Cavazza A, Sturm N, *et al*. Pulmonary carcinomas with pleomorphic, sarcomatoid, or sarcomatous elements: a clinicopathologic and immunohistochemical study of 75 cases. *Am J Surg Pathol* 2003;27:311–24.
3 Mochizuki T, Ishii G, Nagai K, *et al*. Pleomorphic carcinoma of the lung. Clinicopathologic characteristics of 70 cases. *Am J Surg Pathol* 2008;32:1727–35.
4 Huang SY, Shen SJ, Li XY. Pulmonary sarcomatoid carcinoma: a clinicopathologic study and prognostic analysis of 51 cases. *World J Surg Oncol* 2013;11:252.
5 Martin LW, Correa AM, Ordonez NG, *et al*. Sarcomatoid carcinoma of the lung: a predictor of poor prognosis. *Ann Thorac Surg* 2007;84:973–81.
6 Sakata S, Sasaki J, Tanaka R, *et al*. [A case of stage IIIB pulmonary pleomorphic carcinoma that responded well to chemoradiotherapy and surgery]. *Nihon Kokyuki Gakkai Zasshi* 2011;49:131–4.
7 Fishback NF, Travis WD, Moran CA, *et al*. Pleomorphic (spindle/giant cell) carcinoma of the lung. A clinicopathologic correlation of 78 cases. *Cancer* 1994;73:2936–45.
8 Wellmann KF, Chafiian Y, Edelman E. Small bowel perforation from solitary metastasis of clinically undetected pulmonary giant cell carcinoma. *Am J Gastroenterol* 1969;51:145–50.
9 Kagohashi K, Kadono K, Satoh H, *et al*. Intussusception due to intestinal metastasis from lung cancer. *Lung Cancer* 2007;57:247–8.
10 Otera H, Ikeda F, Nakagawa S, *et al*. Intussusception of small intestine due to metastasis of large cell carcinoma of the lung with a rhabdoid phenotype. *Eur Respir Rev* 2010;19:248–52.
11 Jarmin R, Azman A, Rahim R, *et al*. A rare case of intussusception associated with metastasized small cell carcinoma of lung. *Acta Med Iran* 2012;50:782–4.
12 Mandeville Y, de Gheldere C, Vanclooster P. Small bowel intussusception caused by multiple intestinal metastases from a giant cell carcinoma of the lung: a case report. *Acta Chir Belg* 2015;115:49–51.
13 Pollheimer MJ, Eberl T, Baumgartner K, *et al*. Ileocecal intussusception caused by lung cancer metastasis. *Wien Klin Wochenschr* 2009;121:413–16.
14 Shi B, Gaebelein G, Hildebrandt B, *et al*. Adult jejunojejunal intussusception caused by metastasized pleomorphic carcinoma of the lung: report of a case. *Surg Today* 2009;39:984–9.
15 Wou C, Sharp E, Johnson L. Intussusception secondary to a solitary peritoneal lung metastasis. *BMJ Case Rep* 2013. doi:10.1136/bcr-2012-008370
16 Katz ME, Hahn IS, Job GG, *et al*. Intussusception as an unusual complication of metastatic lung cancer. *Conn Med* 1981;45:220–2.
17 Listrom MB, Davis M, Lowry S, *et al*. Intussusception secondary to squamous carcinoma of the lung. *Gastrointest Radiol* 1988;13:224–6.
18 Eng J, Sabanathan S. Carcinosarcoma of the lung with gastrointestinal metastasis. Case report. *Scand J Thorac Cardiovasc Surg* 1992;26:161–2.
19 Issa K, Mullen KD. Large-cell carcinoma of the lung. With major bleeding and intussusception. *J Clin Gastroenterol* 1992;15:142–5.
20 Testini M, Trabucco S, Di Venere B, *et al*. Ileal intussusception due to intestinal metastases from primary malignant melanoma of the lung. *Am Surg* 2002;68:377–9.
21 Yang CJ, Hwang JJ, Kang WY, *et al*. Gastro-intestinal metastasis of primary lung carcinoma: clinical presentations and outcome. *Lung Cancer* 2006;54:319–23.
22 Alvarez Laso C, Paredes Aracil E, Azcano González E, *et al*. Small bowel occlusion due to intussusception of a single metastasis of lung cancer. *Clin Transl Oncol* 2006;8:833–4.
23 Goh BK, Yeo AW, Koong HN, *et al*. Laparotomy for acute complications of gastrointestinal metastases from lung cancer: is it a worthwhile or futile effort? *Surg Today* 2007;37:370–4.
24 Chiu WK, Lin YC, Wang LT, *et al*. Jejunojejunal intussusception secondary to metastasis from adenocarcinoma of the lung—a case report. *Acta Chir Belg* 2009;109:519–22.
25 Kini S, Kapadia RM, Amarapurkar A. Intussusception due to intestinal metastasis from lung cancer. *Indian J Pathol Microbiol* 2010;53:141–3.

26 Lee JW, Kim SK, Park JW, *et al*. Unexpected small bowel intussusception caused by lung cancer metastasis on 18F-fluorodeoxyglucose PET-CT. *Ann Thorac Surg* 2010;90:2037–9.
27 Rashid S, Rajan D, Jacob R, *et al*. Colonic metastases from pleomorphic carcinoma of the lung presenting as an ileocecal intussusception. *ISRN Gastroenterol* 2011;2011:137139.
28 Guner A, Karyagar S, Livaoglu A, *et al*. Small bowel intussusception due to metastasized sarcomatoid carcinoma of the lung: a rare cause of intestinal obstruction in adults. *Case Rep Surg* 2012;2012:962683.
29 Lin MW, Wu CT, Chang YL. Intussusception caused by intestinal metastasis from lung pleomorphic carcinoma. *Ann Thorac Cardiovasc Surg* 2014;20:635–8.
30 Jung SP, Han KM, Bae JW, *et al*. Recurrent intussusception caused by intestinal metastasis of pulmonary pleomorphic carcinoma. *Kaohsiung J Med Sci* 2014;30:378–9.
31 Shoji F, Maruyama R, Okamoto T, *et al*. Long-term survival after an aggressive surgical resection and chemotherapy for stage IV pulmonary giant cell carcinoma. *World J Surg Oncol* 2005;3:32.