Intestinal obstruction caused by giant ileal hamartoma: a case report

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Abstract: Adult intestinal hamartomas is uncommon, intussusception caused by simple intestinal hamartomas are extremely rare. However, there is no report yet like our isolated giant ileum hamartoma. We report an unusual case of a 34-year-old woman who suffered abdominal pain for more than 1 year, and accompanied with obvious symptoms of anemia. The admission CT examination revealed small bowel intussusception. So we had a laparotomy for her. After the reduction of the intussusception, we found a huge mass of 7.5 cm × 2 cm × 2 cm in the intestine, and postoperative pathology showed ileum hamartoma.

Keywords: Ileum; hamartoma; intussusception

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Instruction

Hamartoma is the formation of embryonic tissue deformity or tissue that caused by tumor like hyperplasia (1,2). And it often appears in the lung, kidney, liver and other parts. Occurred in gastrointestinal tract is very rare (3). Although not a true tumor, but it may be malignant, once discovered, needs operation (4,5). The clinical presentation is related to the growth site, in this paper we report the case of an intestinal obstruction caused by giant ileal myoepithelial hamartoma, with symptoms of abdominal pain and chronic anemia.

Case presentation

A 34-year-old woman with abdominal pain admitted to our hospital. Physical examination showed: a temperature of 37.4 ℃, on palpitation of the abdomen, we could touch a mass (12 cm × 3 cm) like a sausage. Laboratory report showed: white blood cell count 9.10×10^9/L; erythrocyte 355×10^{12}/L; hemoglobin 77.0 g/L; platelet count 212×10^9/L; neutrophil 76.2%; tumor associated antigens were normal, stool occult blood positive. Plain abdominal X-ray films revealed incomplete small bowel obstruction. On full abdomen CT scan, a mass was diagnosed involving the mesentery in close proximity with a loop of small intestine with thickened wall, revealing an ileal intussusception (*Figure 1*).

We had a laparotomy for her. There was no ascites in the abdominal cavity, and the intussusception was clearly seen in the ileum. The mass seen was 7 cm × 2 cm in diameter, and closed to ileocecal junction. The intussusception was slightly edematous, no signs of necrosis. We took the lesion part of the intestine out of the abdominal cavity and placed a wet gauze around it, then resented it gently (*Figure 2A*). Made an incision and took the mass out. We found its basilar was wide. Excised the tumor and resected portion of the small bowel making a "V" shape loop, which was sent for pathological examination.

Postoperative gross specimen showed: a tube length 6 cm, diameter 2.5 cm, from the one end of the 3 cm and the other end of 3 cm, seen a 7.5 cm × 2 cm × 2 cm giant mass (*Figure 2B*). The pathology report showed nodular masses in the muscular layer, including a well

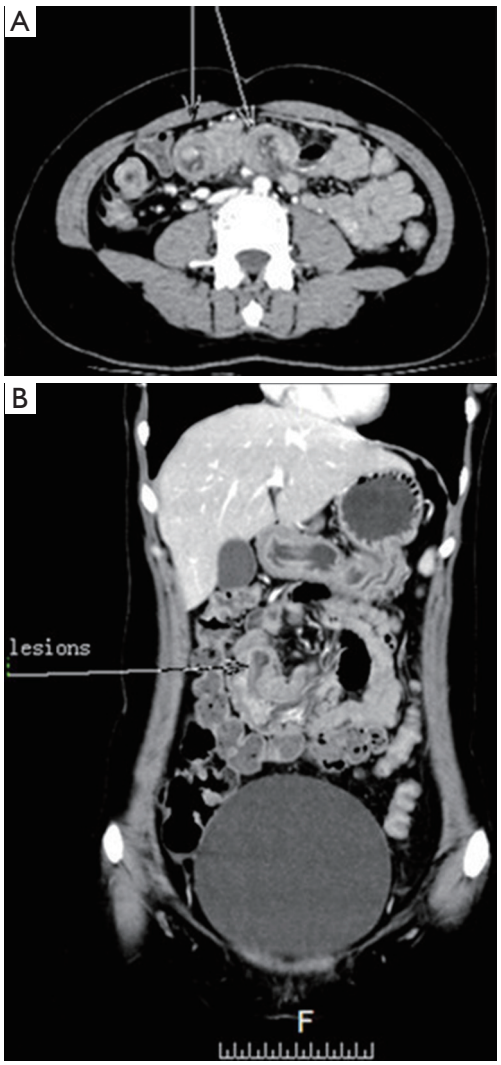

Figure 1 CT scan. (A) (plain): focus on different levels of performance, arrow points to "a concentric circle syndrome", partial wall thickening. The vascular and ring enhancement in the outer tube is enhanced, and the enhanced scanning is layered enhancing; (B) (coronal position): there is a tumor in the intestinal tract, its diameter is about 2 cm. Note: white arrow refers to the lesion.

differentiated fat, blood vessels and ducts. It indicated the change of hamartoma, H&E staining ×100 (*Figure 3A*). Immunohistochemisty: CK19 (+), SMA (+), CD34 (+), S-100 (+), Ki-67 (cell proliferation index <10%), (*Figure 3B-F*).

Discussion

The etiology of hamartoma is unclear. Most scholars believe that it's not a true neoplasm, but in the process of growth it will develop to a true neoplasm, that is, malignant potential (4), once discovered, need operation (3). We have reviewed the literature carefully and found that all age groups can be the disease, but the incidence of young women is relatively high (*Table 1*). The clinical manifestations of ileal hamartoma are abdominal pain, abdominal mass, gastrointestinal hemorrhage, intussusception, intestinal obstruction and corresponding clinical symptoms. Individual patients without any clinical manifestations, only found in physical examination (5).

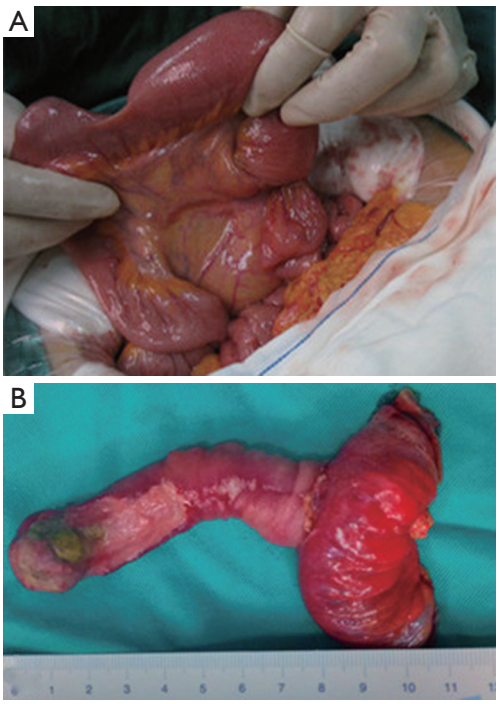

Figure 2 Operation. (A) (Intussusception): intestinal masses caused intussusception; (B) long diameter of the mass is 7.5 cm, and the head has a giant ulcer about 4.5 cm × 2.5 cm; the diameter of the tumor was 2 cm.

On the growth of ileal hamartoma, there are no research reports. Review of previous reports (*Table 1*) of the ileum hamartoma, the maximum was 2.0 cm × 2.0 cm × 1.6 cm. However, in this case report, the hamartoma was 7.5 cm × 2 cm × 2 cm (2), growing along the ileum to distal, and it had a wide basilar. Excepted the basilar, the tumor was not associated with the wall of the tube. A 34-year-old woman with abdominal pain admitted for 1 year to our hospital, self-tolerance, the patient did not visit other hospital before. Asked the patient's living habits, we found that she was enjoying in eating hot, sour and spicy food. So it

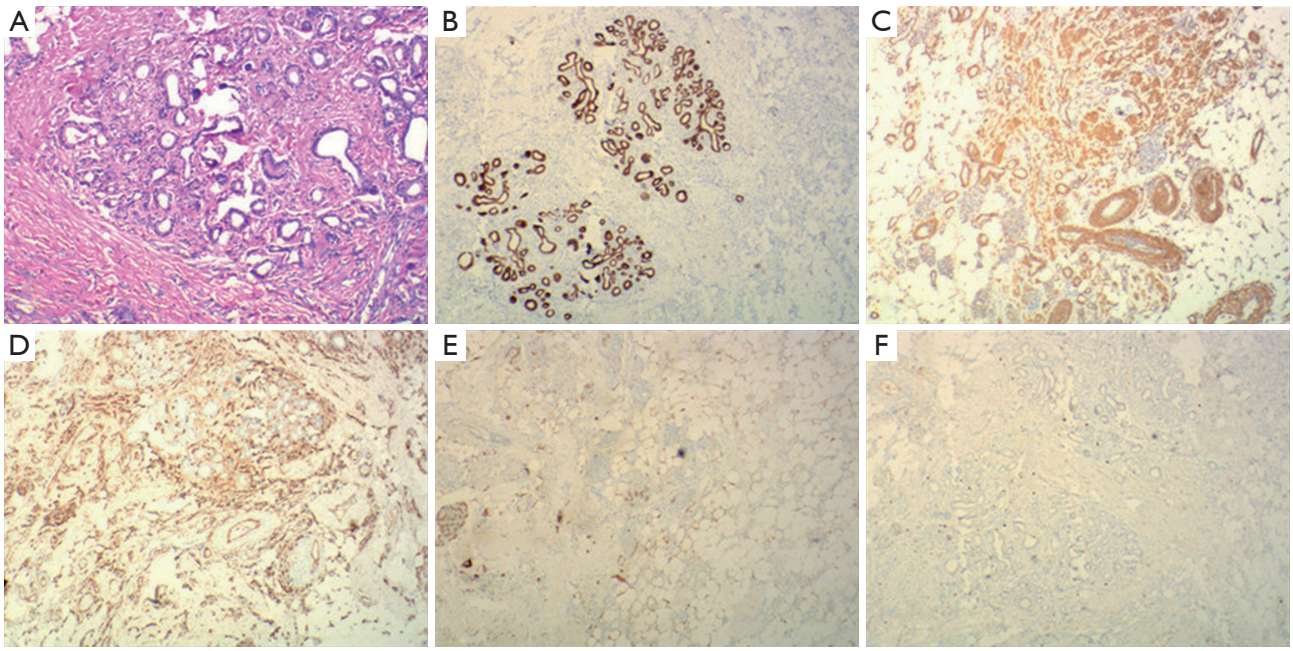

Figure 3 Pathology (H&E staining, ×100). (A) We can find nodular masses in the muscular layer, including a well differentiated fat, blood vessels and ducts; (B) CK19, epithelial tissue (+); (C) SMA, smooth muscle (+); (D) CD34, blood vessel (+); (E) S-100, adipose tissue (+); (F) Ki-67, cell proliferation index <10%. These features indicate that the organization has no atypia, it is the change of myoepithelial hamartoma.

Table1 The reported small intestine (ileum) hamartoma statistics

Case	Authors	Age (sex)	Tumor size (cm^3)	Clinical symptoms	Publication years
1	Tanaka *et al*. (1)	24 years (M)	1.2×1.2×1.0	Melena, pain	1996
2	Ikegami *et al*. (2)	5 months (F)	1.5×1.2×1.0	Vomiting	2006
3	Schwartz *et al*. (6)	8 months (M)	2.0×1.5×1.5	Vomiting	1958
4	Gal *et al*. (7)	82 years (F)	2.0×2.0×1.5	Vomiting, pain	1986
5	Chan *et al*. (8)	5 months (F)	0.8×0.8×0.5	Pain	1994
6	Serour *et al*. (9)	3 years (M)	2.0×2.0×1.6	Vomiting, pain	1994
7	Chen *et al*. (5)	22 years (F)	1.3×1.3×1.0	Bleeding	2015
8	Ours	34 years (F)	7.5×2.0×2.0	Intussusception, pain	2015

can infer that tumor can continue to grow, after receiving stimulation. The patient has chronic anemia symptoms, tumor specimen in combination, the head of it has a giant ulcer about 4.5 cm × 2.5 cm, and it can be judged that that is the cause of anemia. Such a huge tumor, it's moving with the small intestine creeping wave, and it could cause intussusception anytime. If not timely surgical reduction, with the evolution of the disease, it can lead to intestinal obstruction complicated by intestinal necrosis. So, early operation is the best way to cure the ileum hamartoma.

Finally, the small intestine (ileum) hamartoma can continue to grow, after receiving stimulation, further more, broken and causing bleeding. It's moving with the small intestine creeping wave, and it could cause intussusception anytime, and then the occurrence of intestinal obstruction. Therefore, we should pay more attention to the disease and its complications, and to further understand its etiology and pathology, so as to explore and search for better diagnosis and treatment.

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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