Case report

Giant cystic lymphangioma of right mesocolon: A case report

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Abstract

Introduction: Cystic lymphangiomas are rare benign tumors of the lymph vessels and are usually found in children. However, abdominal cystic lymphangioma in mesocolon is extremely rare in adult patients.

Presentation of case: We reported a 15-year-old female with giant cystic lymphangioma of the right mesocolon. On examination, only abdominal pain was confirmed. Abdominal computed tomography (CT) showed a large multiseptated cystic mass. The patient underwent a total right mesocolic excision with the lesion. The patient recovered well on postoperative follow-up and was discharged on the fifth day. No evidence of recurrence had also been found in three months follow-up period.

Discussion: The diagnosis of intra-abdominal cystic lymphoma is often dismissed because the clinical symptoms are nonspecific. It is easy confusion because the ultrasound and CT scan images are relatively similar to the mesenteric and omental cysts. sclerosing therapies may cause long-term consequences such as local recurrences with a very high proportion. Complete resection, including resection of the involved organs, is necessary. With tumors surrounding the colon, surgeons should consider performing removal block colon-lesion.

Conclusion: Complete tumor removal is the optimal choice for the management of intra-abdominal cystic lymphangioma. However, incomplete resection may lead to local recurrence.

1. Introduction

Cystic lymphangioma is a benign tumor of the lymphatic system, mainly affecting the craniofacial region, neck, or chest, common in childhood. Cystic lymphangioma is extremely rare in the adult population, with about 1 in 250,000 hospital admission [1,2]. In the intraperitoneal cavity, cystic lymphangioma was found in 70% of the small mesenteric intestine and the other in the greater omentum, mesocolon, and retroperitoneum [3]. The diagnosis is still challenging due to confusion with mesenteric cysts. We describe a case of giant cystic lymphangioma of mesocolon that was successfully performed tumor removal. This case report has been reported in line with the SCARE criteria [4].

2. Presentation of case

A 15-year-old female (BMI: 16.6 kg/m²) presented to our hospital with 3 months of abdominal pain. She denied vomiting, nausea, and a history of any other abdominal disorder. Physical examination revealed a palpable tumor from the epigastriac to hypogastric region, especially on the right side of the umbilicus. There were unremarkable in routine laboratory and hematologic investigations. Computed tomography showed a large cystic mass with 16.0 × 18.2 cm in size, extending from the lower of the liver to the ileocecal corner, suspecting of a mesenteric cyst or omental lymphangioma (Fig. 1).

During exploration surgery, a huge cystic mass was found covering the right mesocolon (Fig. 2). The surface of the mass showed fibrotic capsules with thick walls. Because of the inability to separate the descending colon and mesocolon from the lesion, total right mesocolic excision with the mass was performed simultaneously. A side-to-side anastomosis was constructed between the terminal ileum and the transverse colon using a linear cutter stapler. After resection, the cystic tumor was grossly measured at 15.2 × 16.8 × 3.2 cm (Fig. 3). The operative time was 254 min, and the blood loss was 350 ml.

Histologically, the cystic wall consisted of fibrous tissue, lymphatic tissue, blood vessels, and smooth muscle, with an inner surface of a single layer of squamous or cuboidal epithelium and no sign of malignancy (Fig. 4). Therefore, the final diagnosis was cystic lymphangioma.

The patient recovered well on postoperative follow-up and was discharged on the fifth day. No evidence of recurrence was also found during the three months follow-up.

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Fig. 1. Computed tomography showed a large cystic mass.

Fig. 2. Intraoperative view remove of a giant cystic lymphangioma.

Fig. 3. Gross specimen showed a cystic lymphangioma with multiseptated.
3. Discussion

Cystic lymphangiomas are rare benign tumors that are found predominantly in children. They generally occur in the neck (75%) and the axillary region (20%). Intra-abdominal lymphoma comprises less than 1%, with the most common location in the small bowel mesentery (70%) and extremely rare in the mesocolon [3,5].

The cause of lymphangioma is still unknown. However, the mechanism of inflammation and fibrosis was accepted by most researchers. No malignant transformation of lymphangioma has been introduced in the literature [6,7].

The clinical characteristics of intra-abdominal lymphangiomas are variable. Abdominal pain and a palpable mass are the common symptoms. Several complications can cause the acute condition of the patient, including a hemorrhaging cyst, a secondary infection, obstruction, or anemia [8,9]. Spontaneous rupture of cysts, gastrointestinal bleeding, and cystic peritoneal lymphangiomatosis is rare presented [9,10].

The diagnosis of intra-abdominal cystic lymphoma is quite difficult because the clinical symptoms are nonspecific. Allen reported a rate of 40% asymptomatic patients in his study and was discovered incidentally on an imaging exam. Even when the patient underwent an ultrasound and CT scan of the abdomen, confusion could still occur because the images are relatively similar to the mesenteric and omental cysts [2]. Abdominal magnetic resonance imaging (MRI) can show better clarifies the nature of the cystic contents. However, it is unable to determine the origin of the tumor [11].

For non-surgical treatment, some sclerosing therapies using Bleomycin, Tissucol, OK-432, and Ethibloc were reported as initial treatments of cystic lymphangiomas. These can reduce the size of the mass. Consequently, these can minimize the frequency and extent of surgical intervention [12–14]. However, Alquahtani et al. have demonstrated that these therapies may cause long-term consequences, such as local recurrences with a very high proportion, up to 100% [15]. Therefore, the optimal choice for intra-abdominal lymphangiomas management is surgery to remove tumors. In most cases of small cysts, total excision without affecting other organs is a priority.

For tumors in solid organs including the spleen, pancreas, liver, or adrenal gland, resection of the lesion along with involved parenchyma should be considered. For cystic lymphangiomas of the mesentery (mesocolon and mesentery), total surgical resection of the tumor with or without bowel can be performed [16]. In order to avoid complications of lymphatic leakage following surgery, a careful examination of the intraoperative lymphatic system is necessary. In this case, we used Ligasure to seal lymphatic ducts and small vessels. After incomplete resection, a recurrence rate of up to 40% was reported. In microscopic residue, the recurrence rate ranges from 5% to 10% [15,16].

4. Conclusion

Intra-abdominal cystic lymphangiomas are rare benign tumors that present a diagnostic challenge of diagnosis. Accurate diagnosis is usually based on histopathology. Abdominal ultrasound and CT scan show a fairly clear cystic mass. Complete resection of the tumor is the optimal choice for lymphangioma management.

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Ethical approval

The study was approved by the research committee, 108 Military Central Hospital, Hanoi, Vietnam.

Consent

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

Registration of research studies

Not applicable for a case report.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Author contribution

1. Nguyen Anh Tuan: performing the operations, manuscript drafting.
2. Nguyen Van Du: follow-up and post-operative management, manuscript drafting.
3. Pham Van Hiep: performing the operations.

Guarantor

Nguyen Van Du.

Declaration of competing interest

Dr. Nguyen Anh Tuan, Dr. Nguyen Van Du and Dr. Pham Van Hiep
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References

[1] S. Kumar, N. Agrawal, R. Khanna, A.K. Khanna, Giant lymphatic cyst of omentum: a case report, Cases J. 2 (2009) 23.
[2] J.G. Allen, T.S. Riall, J.L. Cameron, F.B. Askin, R.H. Hruban, K.A. Campbell, Abdominal lymphangiomas in adults, J. Gastrointest. Surg. 10 (5) (2006) 746–751.
[3] J.H. Kim, W.S. Ryu, B.W. Min, T.J. Song, G.S. Son, S.J. Kim, et al., Acquired omental cystic lymphangioma after subtotal gastrectomy: a case report, J. Korean Med. Sci. 24 (6) (2009) 1212–1215.
[4] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, The SCARE 2020 guideline: updating consensus surgical Case RePoRT (SCARE) guidelines, Int. J. Surg. (London, England). 84 (2020) 226–230.
[5] J.K. Limdi, S. Mehdi, M. Sapundzieski, M. Manu, A.M. Abbasi, Cystic lymphangioma of the mesocolon, J. Gastrointest. Surg. 14 (9) (2010) 1459–1461.
[6] J.L. Hornick, C.D. Fletcher, Intraabdominal cystic lymphangiomas obscured by marked superimposed reactive changes: clinicopathological analysis of a series, Hum. Pathol. 36 (4) (2005) 426–432.
[7] T. Tsukamoto, S. Tanaka, T. Yamamoto, E. Kakinoki, A. Uemichi, S. Kubo, et al., Laparoscopic excision of a retroperitoneal cystic lymphangioma: report of a case, Surg. Today 33 (2) (2003) 142–144.
[8] T. Akashige, K. Sato, H. Odajima, S. Yamazaki, A case report of recurrent intussusception caused by small bowel lymphangioma in an adult, Int. J. Surg. Case Rep. 75 (2020) 126–130.
[9] L. Lepre, G. Costa, D. Baldini, F. Cortese, A. Saputelli, A. Gioffre, et al., Emergency presentation of cystic lymphangioma of the colon: a case report and literature review, Int. J. Surg. Case Rep. 24 (2016) 162–165.
[10] T.K. Ha, S.S. Paik, K.G. Lee, Cystic lymphangioma arising from mesocolon, Clinical gastroenterology and hepatology : the official clinical practice journal of the American Gastroenterological Association. 7 (3) (2009) e14–e15.
[11] H. Kitamura, D. Yamamoto, S. Kadoya, H. Bando, Y. Okayama, H. Minato, Jejunal mesenteric lymphangioma treated by pancreaticoduodenectomy: a case report, Int. J. Surg. Case Rep. 77 (2020) 165–169.
[12] N. Hall, N. Ade-Ajayi, C. Brewis, D.J. Roebuck, E.M. Kieby, D.P. Drake, et al., Is intralesional injection of OK-432 effective in the treatment of lymphangioma in children? Surgery 133 (3) (2003) 238–242.
[13] A. Okada, A. Kubota, M. Fukuzawa, K. Imura, S. Kamata, Injection of bleomycin as a primary therapy of cystic lymphangioma, J. Pediatr. Surg. 27 (4) (1992) 440–443.
[14] V. Martinot, S. Descamps, P. Fèvrier, P. Patenotre, J.M. Brevière, F. Piette, et al., Evaluation of the treatment of cystic lymphangioma by percutaneous injection of ethibloc in 20 patients, Archives de pediatrie : organe officiel de la Societe francaise de pediatrie. 4 (1) (1997) 8–14.
[15] A. Alqahtani, L.T. Nguyen, H. Flageole, K. Shaw, J.M. Laberge, 25 years’ experience with lymphangiomas in children, J. Pediatr. Surg. 34 (7) (1999) 1164–1168.
[16] A. Makni, F. Chebbi, F. Fetirich, R. Ksantini, H. Bediou, M. Jouini, et al., Surgical Management of Intra-Abdominal Cystic Lymphangioma. report of 20 cases, World J. Surg. 36 (5) (2012) 1037–1043.