International Journal of Contemporary Pediatrics
Glutkin AV et al. Int J Contemp Pediatr. 2023 Sep;10(9):1447-1451
http://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20232592

Case Report

Interesting surgical cases of mesenteric cystic lymphangioma

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Received: 11 July 2023
Revised: 09 August 2023
Accepted: 11 August 2023

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ABSTRACT

Mesenteric cystic lymphangioma involving the gastrointestinal tract is uncommon in infants and children. It is a benign tumor due to abnormal formation lymphatic system. These tumors exhibit symptoms varying in site and size of localization. Usually, symptoms are manifested by acute abdominal pain, intestinal obstruction, rising body temperature, and diarrhea. Lymphangioma can easily misdiagnose with other conditions like intestinal obstruction, acute appendicitis, or ovarian cyst in a female child. Therapeutic interventions such as fibrin glue, steroids, bleomycin, OK-432, and laparoscopic-assisted surgery are available. From 2018 to 2022, the Regional Paediatrics Hospital of Grodno, Belarus has recorded two cases of mesenteric lymphangioma. Both were emergency cases. In this scientific paper, we have described a surgical approach with improved outcomes. In both cases, surgical resection of lymphangioma that was rooted alongside the ileum was done. We used laparotomy in our surgical procedure, however, the approach was different, one being lower median laparotomy and another being a transrectal approach. Post-operatively patients were in satisfactory condition. Even though mesenteric lymphangioma is a benign condition, it should be immediately diagnosed and surgically managed to avoid morbidity, and mortality due to an increase in size that can fatally compress the adjacent organs impairing the function of an organ and complications like intestinal obstruction and bleeding. Our approaches with case-1 and case-2, both resulted in effective surgical and post-surgical outcomes. But more data is required to make a population-based influence depending on our approach.

Keywords: Mesenteric cystic lymphangioma, Paediatric, Abdominal ultrasound, Laparotomy

INTRODUCTION

Mesenteric cystic lymphangiomas in the pediatric population are unusual.[1] Mesenteric lymphangiomas originates from lymphatic vessels. Although the cause of these disorders is uncertain, developmental defects that hinder lymphatic vessels from communicating may play a significant role.[2] Lymphangioma does not appear to be a true lymphatic tumor, but rather the result of congenital abnormality of lymphatic vessels. However, some evidence suggests that tumor development might influence by lymphatic blockage, inflammation, abdominal trauma, abdominal surgery, or radiation.[3] It has been hypothesized that mesenteric cystic lymphangioma may develop under these conditions due to the combination of malrotation and intermittent volvulus.[4]

It is a benign tumor with an incidence recorded at 1 in 250,000.[1] Lymphangiomas can be classified into simple, cavernous, and cystic. The simple type is formed by small thin-walled lymphatic vessels and is located superficially on the skin. The cavernous type interconnects to voids of various normal adjacent lymphatics and is formed up of dilated lymphatic vessels and lymphoid stroma. Additionally, the cystic type is composed of lymphatic voids of varying sizes that include collagen and smooth muscle fascicles but have no relation to normal adjacent lymphatics. Nevertheless, the cystic form may also contain

cavernous elements, cystic lymphangioma is not usually easily distinguished from the cavernous type.[5]

Clinically from being asymptomatic to manifesting as severe painful stomach cramps, mesenteric lymphangiomas manifest in a wide range of clinical ways. Additionally, a potentially fatal complication can be the rupture, resulting in anemia as a consequence of intraabdominal or intracavitary hemorrhage, inducing ischemic tissue necrosis, and intestinal gangrene.[6]

The histological analysis shows an abundant lymphatic lymphocyte, lymphoid aggregates, dilated lymphatic lacunae, irregular cysts with walls composed of smooth muscle cells and fibrocytes, mucosa shows foci of microabscess within the crypts, subserous multiloculated cystic lesion partially lined by flat cells without atypia that might be consistent with endothelium, and cysts presenting erosion/ulceration surrounded by granulation tissue and mixed inflammatory infiltrate.[7,8] While laboratory tests reveal leukocytosis, anemia, and an increased C-reactive protein.[9,10]

Hence, in our cases, we have described clinical manifestation, confirmatory diagnosis, and surgical treatment of the lymphangioma cases presented at the Regional Paediatrics Hospital of Grodno, Belarus. Both patients presented with signs of intestinal obstruction, loosening of the stools, a rise in body temperature to a febrile number, acute colicky pain, and recurrent vomiting in the state of emergency.

CASE REPORT

Case 1

A 2-year-old child presenting in Central District Hospital of Grodno region. Initially, he was noticed to have symptoms of intestinal infection like colicky abdominal pain, repeated vomiting, an increase in body temperature to febrile number, and loosening of stool. During an objective examination, the child had pain in all the quadrants. Further investigation of abdominal ultrasound was done and it showed signs of intestinal obstruction. There was an anechoic-like single pedunculated mass with a fluid-filled content and the loops were distinctly dilated.

Later the child was transferred to the Regional Paediatrics Hospital for further treatment. After the clinical sign of intestinal obstruction appeared, the child was taken to the operating room for exploratory laparoscopy. In laparoscopy, it was visualized that the child had dilated loops of the small intestine and in the right half of the abdomen there was a formation of about 15 cm diameter of soft elastic consistency in the mesentery.

In this child, the pubic symphysis and umbilicus were taken as the standard upper and lower midline laparotomy incision boundaries, respectively. An eventration of the conglomerate was adjacently formed on the terminal ileum until the ileocaecal angle rooted mainly in the thickness of the mesentery of the small intestine over the distance of 25-30 cm. A cyst on the mesentery of the ileum was projecting in 5-6 cm diameter and underlying up to 2-2.5 cm in diameter, hence squeezing the lumen of the ileum as shown in (Figure 1). The tumor was resected within the margins of healthy tissue and an ‘end to end’ ileo-ileo anastomosis with two-row sutures was performed. Simultaneously appendectomy was performed.

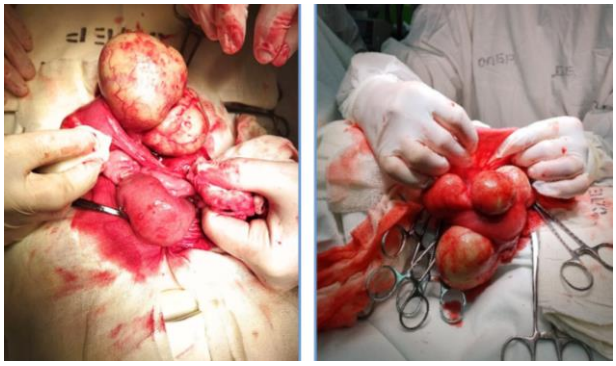

Figure 1: Visualization of mesenteric lymphangioma at laparotomy in a 2-year-old child.

Subsequently, a pedunculated mass was sent for a histopathological examination that showed edema, tortuous blood vessels on the wall, hyperplasia of the lymphoid tissue in the submucosal layer, purulent inflammation in the serosa layer, the infiltrate penetrating the muscular fold of the small intestine. The mass was completely covered with fibromuscular tissue, along with a full-length capillary-cavernous hemangioma and severe inflammation. The analysis discussed above led us to make the diagnosis of a benign tumor as lymphangioma.

Case 2

A 5-year-old child presenting in Central District Hospital of Grodno region. At first, his symptoms of an intestinal infection were frequent episodes of vomiting, excruciating stomach pain, a rise in body temperature to a febrile level, and loosening of the stools. During an objective examination on admission, a painless soft-elastic mass was palpated on the right umbilical area.

On abdominal ultrasound, a 20×8 cm anechoic single pedunculated mass with a fluid-filled cavity and severely enlarged loops was visible. In the small pelvis, a bilocular cyst was seen as shown in (Figure 2).

The child was transferred to the Regional Paediatrics Hospital for further treatment. Following clinical sign of acute intestinal obstruction was seen and the child was taken to the operating room for an exploratory laparoscopy, which revealed the mesenteric white mass enveloping on 3rd part of the ileum it turned out to be a trilocular cyst formed of about 20×10 cm.

In the child, a transrectal laparotomy was done. The cyst was located at a distance of two meters from the ileo caecal angle as shown in (Figure 3). The trilocular cyst was fluid filled and squeezing the lumen. The cyst was then punctured and about 500 ml of fluid was aspirated. The resection was done where the cyst was present within the margins of healthy tissue, carried out by 'end to end' ileo-ileo anastomosis with two-row sutures was done.

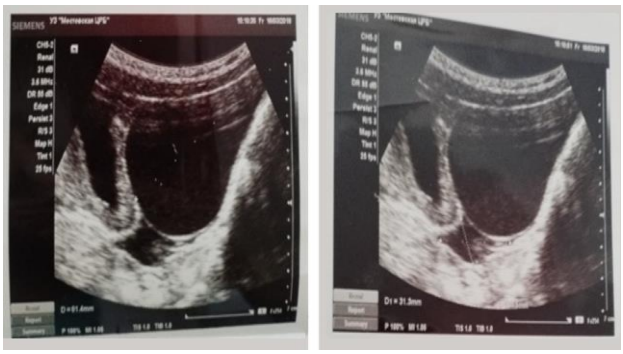

Figure 2: A bilocular cyst on abdominal ultrasound of a 5-year-old child.

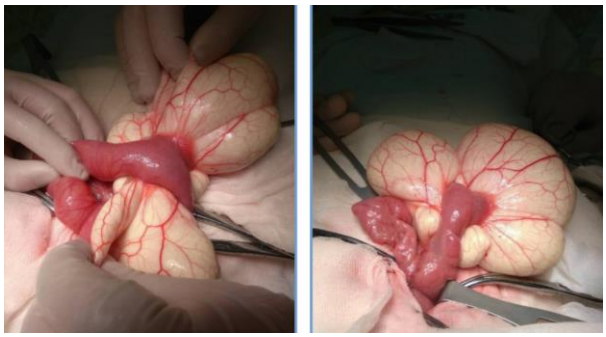

Figure 3: Visualization of mesenteric lymphangioma at laparotomy in a 5-year-old child.

The resected part was sent for a histopathological examination which showed edema of the submucosal layer and an adjacent fibrous wall had no epithelial lining. No fistula had formed between the cyst and intestine. Aspirated fluid was sent for biochemical analysis and the result showed (a Rivalta test positive which means the presence of exudate fluid, pH 7, relative density -1035, total protein -180 g/l, glucose 5.8 mmol/l, 40% neutrophils, eosinophils 2%, lymphocytes 58%) under the microscope.

Based on these analyses the diagnosis of a benign tumor as lymphangioma was made.

In case 1, the child was presenting with severe abdominal pain, loosening of stools, and an increase in temperature to a febrile number. Abdominal ultrasound showed a pedunculated mass. Immediately after admission to the department, the child was rushed to the operating room for an exploratory laparoscopy that showed a cyst in mesentery in the right half of the abdomen. Following exploratory laparoscopy, a lower median laparotomy was done and excised the tumor within the healthy margin of the tissue. Post-operatively there were no complications and the outcome was satisfactory. The patient was discharged from the hospital on the 10th day after the operation. Four months of follow-up visits showed no complications.

In case 2, the child was presenting with bouts of vomiting, severe abdominal pain, stool loosening, and an increase in temperature to a febrile number. An ultrasound of the abdomen revealed a bilocular pedunculated mass. After being admitted to the department, the child was brought to the operating department for an exploratory laparoscopy. Although it appeared to be a bilocular cyst on ultrasound, an exploratory laparoscopy revealed it to be a trilocular cyst. Following exploratory laparoscopy, transrectal laparotomy was done, and excised tumor within a healthy margin of the tissue. Following surgery, there were no complications, and the result was satisfactory. On the 10th day following the operation, the patient was discharged from the hospital. Six months of follow-up visits showed no complications.

DISCUSSION

Mesenteric cystic lymphangiomas are unusual benign tumors that are frequently found in children. It has varied clinical presentations, from asymptomatic to acute abdominal pain.[1,6] The symptoms that both patients have shown include intestinal obstruction, loosening of the stools, a rise in body temperature to a febrile number, acute colicky pain, and vomiting. The most frequent symptom appears to be abdominal discomfort, which can occasionally mimic acute appendicitis. Other symptoms include Meckel's diverticulitis, constipation, bowel blockage, volvulus, and infarction, mostly when they occur in the mesentery.[7] So, the primary care physician or pediatricians should be aware of the lymphangioma condition as a differential diagnosis. An anechoic pedunculated cyst was visible on the abdominal ultrasound of both patients. A 15 cm diameter white cyst in a 2-year-old child and a 20×10 cm trilocular cyst in a 5-year-old child were both seen using a laparoscopic method for an exploratory diagnosis. The use of laparoscopy is recommended for both diagnostic and therapeutic purposes.[11] The excised tissue from both patients was sent for histopathological examination, and the diagnosis of lymphangioma was made. There are no reports of mesenteric lymphangioma-related malignant tumors in the literature. A Hodgkin lymphoma that originated from a mediastinal cystic lymphangioma has been described in one incidence.[12] Herein, in our cases we have described clinical manifestation, confirmatory diagnosis, and surgical treatment of the lymphangioma cases presented at the Regional Paediatrics Hospital of Grodno, Belarus.

While the lymphangioma can be removed surgically or endoscopically. Endoscopic excision is typically used for lymphangiomas with a maximum diameter of 2 cm or

less.[8] In our cases, it was not possible to use endoscopic excision as the diameter of the cysts was more than 2 cm. The primary treatment for mesenteric cystic lymphangioma is a mass excision performed during an open laparotomy. Sometimes an intestinal resection is necessary for the cyst that is strongly adhered to the intestinal wall.[13] However, surgery with a laparotomy was performed to remove the cyst, in a 2-year-old child, a lower median laparotomy approach was used because the cyst was located in the terminal ileum until the ileocaecal angle, whereas in a 5-year-old child, a transrectal laparotomy approach was used as the cyst was located 2 meters from the ileocaecal angle following an 'end to end' ileo-ileal anastomosis in both cases. Although aspiration and injection of sclerosant drugs may be suggested for an emergency decompression, they have a high recurrence rate when used as definitive therapies. Other therapies including fibrin glue, OK-432, ethibloc, bleomycin, and steroids haven't shown to be more effective than surgery in terms of outcomes.[14] Fewer post-operative ileus, reduced parietal scars, and a lower likelihood of intestinal adhesion with subsequent blockage are all benefits of laparoscopy. Due to the increased risk of extended bowel resection and post-operative chylous ascites, lymphatic malformation getting into the colonic or intestinal mesentery is a contraindication to laparoscopic resection; cases of extensive lymphatic malformations that are closely associated with major blood vessels surgical excision were thought to be difficult and could have caused unneeded tissue damage raising the possibility of conversion, thus laparoscopy should be carefully evaluated as a therapy option.[15]

CONCLUSION

Regional Paediatrics Hospital in Grodno has identified two mesenteric lymphangioma incidences between 2018 and 2022. Similar symptoms, including severe stomach discomfort and clinical indications of intestinal obstruction, were present in both patients. They underwent abdominal ultrasound, which revealed a 20×8 cm pedunculated tumor in a 5-year-old patient and a fluid-filled anechoic single pedunculated mass in a 2-year-old child. Due to the deteriorating condition of both patients, they were rushed for an exploratory laparoscopy. A white soft elastic consistency cyst with a 15 cm diameter was found in a 2-year-old child, whereas a tri-loculated cyst with a 20×10 cm diameter was found during exploratory laparoscopy. Since the symptoms in both cases were getting worse, they were both emergencies, necessitating laparotomies. Both the methods used for laparotomies were different since the cysts were located at various places. After an operation, both patients have been discharged on the 10th day in good health, and no complications have been reported as of now. We, therefore, assume through our scientific investigation that surgical excision followed by an end-to-end anastomosis with two-row sutures through the approach of laparotomy would be the curative approach of choice for mesenteric lymphangioma to achieve a low to zero recurrence rate. To make a population-based influence based on our technique, however, more data is required.

ACKNOWLEDGMENTS

The authors would like to thank patients, their legal guardians and Dr. Mehul Hitesh Sadadiwala.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Glutkin AV, Khartanovich VV, Patel GR. Interesting surgical cases of mesenteric cystic lymphangioma. Int J Contemp Pediatr 2023;10:1447-51.