Pediatr Pol 2021; 96 (4): 285–287
DOI: HTTPS://DOI.ORG/10.5114/POLP.2021.109297
Submitted: 8.05.2021, Accepted: 27.06.2021, Published: 16.09.2021

PEDIATRIA Polska
PISMO POLSKIEGO TOWARZYSTWA PEDIATRYCZNEGO
Polish Journal of PAEDIATRICS
OFFICIAL JOURNAL OF POLISH SOCIETY OF PAEDIATRICS

CASE REPORT

A rare presentation of mesenteric cystic lymphangioma in 5-year-old girl

Patrycja Sosnowska-Sienkiewicz, Cezary Miedziarek, Przemysław Mańkowski

Department of Pediatric Surgery, Traumatology and Urology, Poznan University of Medical Sciences, Poznan, Poland

ABSTRACT

Acute abdominal pain is a frequent event in the paediatric population and appendicitis is the most common cause.

Abdominal lymphangiomas occur most frequently in the mesenterium and are usually asymptomatic. Nevertheless, they can manifest as acute abdomen, needing surgical treatment. We report the case of a 5-year-old female operated on due to suspicion of acute abdomen. The pathological lesion was surgically removed during the second operation.

The differential diagnosis of acute abdomen should be initially focused on the most common abdominal diseases. However, rare cases, like the presented one, must also be taken into consideration. Ultrasound examination plays important role in the initial differentiation of the causes of abdominal pain.

KEY WORDS:

abdominal cystic lymphangioma, acute abdomen, appendicitis, surgery.

INTRODUCTION

Lymphangiomas are rare lesions of the lymphatic vessels. Their aetiology is uncertain. In most cases lymphangiomas are congenital, often associated with genetic disorders (e.g. trisomies 13, 18, and 21, Noonan syndrome, Turner syndrome) [1]. Abdominal lymphangiomas are rare and occur most frequently in the mesenterium. Mesenteric lymphangiomas have varied clinical presentations, from asymptomatic to acute abdominal pain [2]. They can also cause life-threatening complications like rupture, anaemia due to intraabdominal or intracavitary bleeding, ischemic tissue necrosis, intestinal gangrene, intestinal obstruction with the needing for surgical exploration [3]. Based on our experience, we would like to present a clinical case of a 5-year-old female patient whose acute abdominal pain was attributed to appendicitis and solved with two laparotomies.

CASE REPORT

A 5-year-old female was referred to district hospital by the general practitioner because of suspected appendicitis. At arrival to the Emergency Department, the abdomen was tense with muscular defence. Increased inflammatory markers (CRP 220.8 mg/l, white blood cells 19.6 $\times 10^9$/l) and anaemia (haemoglobin 8.6 g/dl, haematocrit 25.7%) were revealed in laboratory tests. The patient's temperature was 36.6°C. The ultrasound examination was not performed at this point. On the basis of physical examination and laboratory tests, the patient was qualified for surgery as a suspicion of appendicitis. During the laparoscopic operation, the conversion to laparotomy was necessary. The appendix was normal, but a pathological mass in the umbilical region was observed. The decision of ending the surgery and transferring the patient to our referral centre was made. The lesion was not typical and surgeons assessed their experience as too small to operate this. On admission, the patient was apathetic. The skin

ADDRESS FOR CORRESPONDENCE:

Patrycja Sosnowska-Sienkiewicz, Department of Pediatric Surgery, Traumatology and Urology, Poznan University of Medical Sciences, Poznan, Polande-mail: patrycja.sosnowska@outlook.com

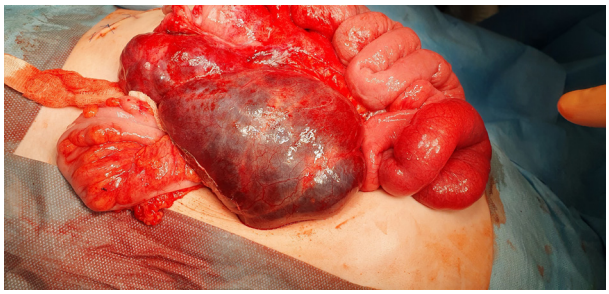

FIGURE 1. Mesenteric lymphangioma filled with lymphatic fluid and blood

was pale with no other changes. Due to strong abdominal defence, the palpation of internal abdominal organs was very difficult. The ultrasound examination and abdominal CT-scan showed a large cystic lesion (7.2 × 13.2 × 12.8 cm) in the umbilical region and retroperitoneal space ascending from the small intestine's mesenterium. The bowel loops were moved upward, the aorta slightly lateral. The inferior vena cava and the left kidney were dislocated. The next explorative laparotomy was performed. A vast cystic lesion ascending from the mesenterium was dissected. Cysts were filled with lymphatic fluid and blood. Free fluid visible in the abdominal cavity contained an admixture of blood. The intraoperative diagnosis was of a mesenteric lymphangioma (Figure 1).

The lesion was radically resected and removed en bloc. Appendectomy was also performed. After a week of observation, the patient was discharged from the hospital with no postoperative complications.

DISCUSSION

Acute abdominal pain, which occurred in our patient, is a frequent ailment in the paediatric population [4]. The literature indicates that in the overall paediatric population, the most common causes are respectively: appendicitis, which was a primary diagnosis in the presented case, bowel obstruction, child abuse, constipation, dietary indiscretions, gallbladder disease, gastroenteritis, haemolytic uremic syndrome, mesenteric adenitis, pancreatitis, sickle cell crisis, trauma, upper respiratory infection, and urinary tract infection [5]. The life-threatening causes of acute abdominal pain, like haemorrhage, obstruction, and perforation of the gastrointestinal tract, deserve special attention and could have been suspected in our patient due to anaemia and increased inflammatory markers at the laboratory tests [6]. It is essential to differentiate between surgical and non-surgical as well as urgent and non-urgent conditions presenting as acute abdominal pain [7]. The differentiation is not simple. The patient's medical history, physical examination, imaging and, laboratory tests are the basis of diagnosis, nevertheless, diagnostic laparoscopy may be occasionally needed in some patients for the final verification as in the presented case [8, 9].

As already mentioned, appendicitis is the most common surgical emergency and a frequent cause of abdominal pain [10]. Many conditions should be differentiated from appendicitis, among them the ones above and others like intussusception, acute cholecystitis, perforated peptic ulcer, mesenteric adenitis, Meckel's diverticulitis, colonic or appendicular diverticulitis, pancreatitis, and rectus sheath hematoma. Urological, gynaecological, and systemic conditions must also be taken into consideration [11]. The described patient's acute abdominal pain, muscular defence and laboratory tests results belong to the symptoms of appendicitis as described in the literature. Other common symptoms and clinical findings in paediatric patients with this condition are nausea, vomiting, fever, diarrhoea, migration of pain, cough, respiratory distress, and leukocytosis with neutrophilia in the blood count [12, 13]. We did not observe such symptoms in our patient.

Mesenteric lymphangioma is a rare finding because it mainly occurs in the head, neck, and axilla [14]. Lymphangiomas in the abdominal cavity are highly uncommon, but if they appear, they originate from the mesenterium [15]. Same as for our patient. Imaging is beneficial in the diagnosis of mesenteric lymphangiomas. Ultrasonography is used for the initial diagnosis of the lesion because it is considered a highly sensitive tool for diagnosing cystic lesions and due to lack of radiation exposure [16, 17] CT-scan provides information on anatomical localization, size, and involved structures, which is crucial to plan the surgical treatment. Therefore it was used in our patient to complete the diagnostic imaging, nevertheless, magnetic resonance imaging is also considered to be an effective examination in these cases [16]. In our case, the symptoms of acute abdomen have evolved due to prolonged intra-cystic haemorrhage, but other reported reasons like torsion or rupture can cause the acute abdominal pain [18]. The treatment of mesenteric lymphangioma in this case was surgical resection due to the patient's clinical condition. The lesion is classified as benign, but it is essential to remove it radically. Lymphangioma tends to recur and infiltrate adjacent structures [19]. Even though the complete surgical resection is the most effective way of treating mesenteric lymphangiomas. A minimally invasive approach using percutaneous drainage and sclerosis with ethyl alcohol was recently reported as innovative but also efficient therapy [20].

Considering the diagnostic criteria proposed in the Appendicitis Inflammatory Score (AIR), the patient at the district hospital received the score that requires additional evaluation using imaging examinations or laparoscopy [21]. Probably an ultrasound examination before the decision about surgery would save the child from the necessity of two laparotomies.

It emphasises the importance of ultrasound examination in the differential diagnosis of abdominal pain. It is suggested that USG could be sufficient for the ini-

tial diagnosis of the lesion in our patient. Unfortunately, the remaining results of both the physical examination and laboratory tests could suggest appendicitis.

CONCLUSIONS

Differential diagnosis of acute abdomen in paediatric patients must include rare conditions, like lymphangiomas. Ultrasound examination plays important role in the initial differentiation of the causes of abdominal pain. Radical surgery is an effective treatment for abdominal lymphangiomas.

DISCLOSURE

The authors declare no conflict of interest.

REFERENCES

1. Miceli A, Stewart KM. Lymphangioma – StatPearls – NCBI Bookshelf. https://www.ncbi.nlm.nih.gov/books/NBK470333/ (accessed Apr. 25, 2021).
2. Katı Ö, Güngör Ş, Kandur Y. Mesenteric cystic lymphangioma: Case report. J Pediatr Surg Case Reports 2018; 35: 26-28.
3. Nagano H, Kimura T, Iida A, et al. Cystic lymphangioma in the peripheral jejunal mesentery in an adult and excision with laparoscopic-assisted surgery: a case report. World J Surg Oncol 2019; 17: 170.
4. D'Agostino J. Common abdominal emergencies in children. Emerg Med Clin North Am 2002; 20: 139-153.
5. Reust CE, Williams A, Acute Abdominal Pain in Children. Am Fam Physician 2016; 93: 830-836.
6. Kim JS. Acute abdominal pain in children. Pediatr Gastroenterol Hepatol Nutr 2013; 16: 219-224.
7. Hijaz NM, Friesen CA. Managing acute abdominal pain in pediatric patients: current perspectives. Pediatric Health Med Ther 2017; 8: 83-91.
8. Bartley MK. Acute abdominal pain. Nurs Crit Care 2007; 2: 38–45.
9. Stefanidis D, Richardson WS, Chang L, et al. The role of diagnostic laparoscopy for acute abdominal conditions: an evidence-based review. Surg Endosc 2009; 23: 16-23.
10. Gadiparthi R, Waseem M. Pediatric Appendicitis – StatPearls – NCBI Bookshelf. https://www.ncbi.nlm.nih.gov/books/NBK441864/ (accessed May 04, 2021).
11. Humes DJ, Simpson J. Acute appendicitis. BMJ 2006; 333: 530-534.
12. Hamid KA, Mohamed MA, Salih A. Acute Appendicitis in Young Children: A Persistent Diagnostic Challenge for Clinicians. Cureus 2018; 10: e2347.
13. Salö M, Friman G, Stenström P, et al. Appendicitis in children: evaluation of the pediatric appendicitis score in younger and older children. Surg Res Pract 2014; 2014: 1–6.
14. Aprea G, Guida F, Canfora A, et al. Mesenteric cystic lymphangioma in adult: a case series and review of the literature. BMC Surg 2013; 13 Suppl 1: A4.
15. Wani I. Mesenteric lymphangioma in adult: a case series with a review of the literature. Dig Dis Sci 2009; 54: 2758-2762.
16. Siddique K, Bhandari S, Basu S. Giant mesenteric lymphangioma: a rare cause of a life-threatening complication in an adult. BMJ Case Rep 2010; 2010: bcr0420102896.
17. Esen K, Özgür A, Karaman Y, et al. Abdominal nonparenchymatous cystic lesions and their mimics in children. Jpn J Radiol 2014; 32: 623-629.
18. Gunadi, Kashogi G, Prasetya D, Fauzi AR, et al. Pediatric patients with mesenteric cystic lymphangioma: A case series. Int J Surg Case Rep 2019; 64: 89-93.
19. Losanoff JE, Richman BW, El-Sherif A, Rider KD, Jones JW. Mesenteric cystic lymphangioma. J Am Coll Surg 2003; 196: 598-603.
20. Pozzi G, Ferrarese A, Borello A, et al. Percutaneous drainage and sclerosis of mesenteric cysts: literature overview and report of an innovative approach. Int J Surg 2014; 12 Suppl 2: S90-S93.
21. Rentea RM, Peter SDS, Snyder CL, Pediatric appendicitis: state of the art review. Pediatr. Surg Int 2017; 33: 269-283.