Case Report

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A Ruptured Sigmoid Mesocolon Hemangioma Presenting as Acute Abdomen: A Case Report and Review

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Abstract

Hemangiomas of the GI tract and mesentery are uncommon benign vascular lesions. While spontaneous bleeding distinguishes the gastrointestinal tumor variant, clinical signs of mesenteric hemangiomas are generally nonspecific. In the majority of cases, despite advances in imaging technology, surgery and histopathological analysis are still used to make final diagnoses. We present a case report of a 13-year-old female patient who was admitted with progressive lower abdominal pain and distension and suffered from persistent abdominal pain for 04 days. An irregular hypo-echoic area noted in left iliac fossa suggestive of bowel mass was detected on the ultrasound scan. Due to sonographic signs of an intraabdominal bowel mass, a diagnostic laparoscopy was performed, which revealed a large ruptured hemangioma originating from the sigmoid mesocolon. Although uncommon, pediatric surgeons should consider gastrointestinal hemangiomas as a possible differential diagnosis for large intraabdominal tumorous masses, particularly in young adults.

Keywords: Hemangioma, Sigmoid mesocolon, Gastrointestinal.

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Introduction

Hemangiomas are benign tumor lesions that are classified as mesodermal vascular hamartomas.[1–5] Hemangiomas of the digestive tract are extremely uncommon. Those of mesenteric origin are even more uncommon.[2] They can appear as single or multiple lesions (hemangiomatosis). In the latter case, a connection to similar neoplasms in other locations is possible, which can be caused by syndromes such as Osler-Weber-Rendu disease, Maffucci syndrome, Klippel-Trénaunay syndrome, or the congenital blue rubber bleb nevus syndrome.[6–8] The symptoms of hemangiomas are determined by where the primary tumor is located. Despite this, the most common primary manifestation is spontaneous bleeding,[9,10] which results in free intraabdominal fluid accumulation (hematoperitoneum).

Complete surgical tumor resection is the gold standard for the treatment of mesenterial hemangiomas. Following the complete removal of this tumor, no recurrences have been reported. Although minimally invasive surgery is preferable to open surgery, tumor bleeding can occur, especially in cases of large intraabdominal masses.

The purpose of this article is to go over the clinical, radiological, and histological characteristics of ruptured mesenteric hemangiomas, which are a possible differential diagnosis for acute abdomen.

Case report

A 13-year old female patient was admitted to the Imperial hospital, Chittagong, Bangladesh, with a progressive abdominal distention and increasing diffuse abdominal pain which was initially mostly on left abdomen with fever. No nausea, vomiting, or any other severe constitutional symptoms were mentioned by the patient. The patient had no history of previous abdominal surgery or trauma and had never been in a hospital before.

Physical examination revealed a moderate distention of the abdomen, with diffuse pain caused by digital pressure, with signs of peritonism. An ill-defined mass was palpable extending to the lower abdomen on left side.

There were no abnormal findings in the routine laboratory tests. Measurement of tumor markers was not done. The ultrasound examination revealed an irregular hypo-echoic area noted in left iliac fossa measuring about 3.4 cm X 1.3 cm with mild ascites which deems suggestive of bowel mass and differential diagnosis was short segment bowel inflammation. An abdominal computerized tomography (CT) scan was suggested by radiologist but not done due to patient financial condition.

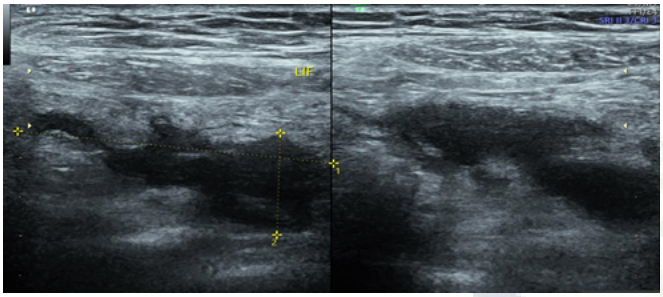

Figure 1: The ultrasound shows an irregular hypo-echoic area noted in left iliac.

Due to deteriorating patient's condition and progression of signs of peritonism, surgical intervention was indicated and a diagnostic laparoscopy was performed. The exploration of the peritoneal cavity revealed an oozy ruptured surface of inner wall of hemangioma originating from the sigmoid mesocolon and hemorrhagic peritoneal fluid which was around 300 ml in amount. There were multiple rounded whitish calcifications seen over the inner wall of ruptured hemangioma. The ruptured hemangioma was excised from the sigmoid mesocolon by ligature and closed with 2/0 vicryl (RB). There were no signs of infiltration of the structures nearby. After removal of the tumor the perfusion of the sigmoid colon seemed normal. Inspection of the sigmoid showed no signs of diminished vascular perfusion such as decreased pulses in the mesosigmoid and macroscopic signs of ischemia (color). The patient was discharged on the third postoperative day.

Histopathological examination of the resected specimen revealed large blood filled vascular spaces within in the fibro fatty tissue of the mesosigmoid and presence of fat necrosis with infiltration of chronic inflammatory cells and foamy macrophages along with fibrosis.

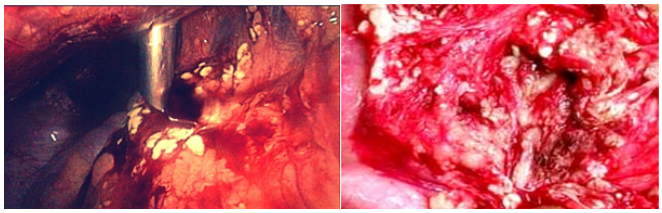

Figure 2: Peroperative images of the ruptured tumorous mass (inner wall) originating from the sigmoid mesocolon showing white round calcifications.

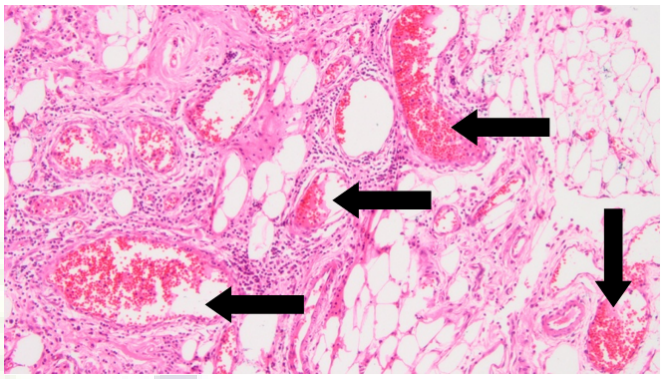

Figure 3: H&E staining of the resected specimen revealed characteristic dilated blood vessels (arrows) (100xmagnification).

Discussion

Hemangiomas are rare hamartomatous lesions that develop from embryonic sequestrations of mesodermal tissue and can affect any organ.[1–5] While intestinal hemangiomas are common and have been documented in the literature, a cavernous hemangioma of the mesenterium is a very uncommon tumor.[2]

Aside from angiomatous lesions in Maffucci syndrome, hemangiomas are typically benign and do not have the potential to become cancerous. Hemangiomas of the gastrointestinal tract can affect people of any age, but they are more prevalent in young adults.[1,3,4] Female sex preference has been discovered.[1,2]

Hemangiomas are soft, compressible bluish purple lesions that histologically consist of vast blood-filled cavities or sinuses lined by single or several layers of endothelial cells. As a result of thrombosis within sinuses, caused by perivascular inflammation and blood flow stasis, degenerative alterations such as hyalinization and localized calcification emerge.[11] Calcification can lead to the formation of phleboliths, which are a diagnostic characteristic detected in 26 to 50 percent of patients.[12]

Despite the absence of phleboliths on preoperative imaging (abdominal radiograph and CT scan) in the case presented, the histology material revealed evidence of perivascular inflammation and calcification close to the normal dilated blood vessels. Various signs and symptoms have been recorded in the literature depending on the tumor's location. 80 percent of patients with gastrointestinal localization have symptoms such as bleeding or mechanical bowel obstruction.[9,10] Hemorrhage associated with cavernous hemangiomas usually occurs suddenly and manifests as hematemesis or melena. Anemia can be the primary symptom in cases of persistent, recurrent bleeding. Polypoid hemangiomas or bowel perforation are rare causes of intussusception.[13,14] Hematoperitoneum can be caused by bleeding from extraluminal hemangiomas, such as those in the mesenterium or omentum. These patients' main complaints are abdominal pain and increasing distention.[13–15] The patient in this case complained of stomach pain and distention. The ultrasound conducted on admission revealed evidence of free intraabdominal fluid. The huge magnitude of the lesion was most likely to blame for the symptoms. Obviously, an intraabdominal hemorrhage produced the rapid abdominal distension (over the course of four days). Histological analysis of the resected specimen verified the latter. However, Kasabach-Merritt syndrome (also known as hemangioma thrombocytopenia syndrome) has been linked to a rare occurrence of a cavernous hemangioma of the gut and mesenterium.[16] Gastrointestinal and mesenteric hemangiomas are frequently misdiagnosed, despite improved imaging techniques. The literature reports an average of 19 years from the onset of symptoms to the ultimate right diagnosis due to unspecific symptoms.[17] The tumor's extent, multiplicity, vascularization, and involvement of the intestine or other intraabdominal or retroperitoneal structures are all revealed by an ultrasound examination, whereas a CT scan provides additional information about the tumor's extent, multiplicity, vascularization, and involvement of the intestine or other intraabdominal or retroperitoneal structures.[18] In cases of acute bleeding, mesenteric angiography can be helpful in determining the exact location of the tumor as well as the possibility of interventional ablation of the arterial input. To be successful, the procedure requires a sufficient amount of bleeding (0.5 mL/min). A selective mesenteric angiography can be useful in detecting the exact location of the bleeding as well as the extent of resection to be conducted in patients with diffuse hemangiomatosis of the mesentery, where a bleeding out of a single vessel cannot be recognized.[19]

The treatment of ruptured mesenteric hemangioma of the gastrointestinal tract or mesentery is either open or laparoscopic surgical excision. A minimally invasive laparoscopic approach could be considered as the gold standard for intraabdominal ruptured hemangioma of unknown origin and entity. Nevertheless, we may prefer open surgery due to the tumor size and bleeding risk. Nonoperative techniques such as sclerotherapy, cryosurgery or interventional angiography usually not recommended in case of rupture hemangioma. Recurrence after complete resection has not been reported.[3,20]

Conclusion

Although extremely rare, hemangiomas of the mesentery should be explored as a differential diagnosis in individuals who have abdominal pain and imaging that shows significant intraabdominal tumorous masses. Despite contemporary radiographic imaging techniques, a soft tissue sarcoma is a more common differential diagnosis, which may lead to a tumor sample. Given the risk of complications from percutaneous hemangioma biopsy, exploratory laparotomy or laparoscopy should be considered if imaging techniques leave room for ambiguity.

Authors' contributions

Adnan Walid wrote the manuscript, with assistance from Raju Das, Effat Sharmin, Adnan Bin Saleh, Arunabha Mallick, Utpol Chowdhury and Mymoon Redwan Chowdhury. Histological staining was done by Tareak Al Nasir, and radiographic imaging was done by Puspita Chowdhury. Utpak Das and Dil Anziz are in charge of assisting with surgery. Adnan Walid performed the patient's surgical treatment. The final manuscript was read and approved by all authors.

References

1. Garvin P, Herrman V, Kaminski D. Benign and malignant tumors of the small intestine. Curr Probl Cancer. 1979;3(9):4–46. Available from: https://doi.org/10.1016/s0147-0272(79)80037-9.
2. Takamura M, Murakami T, Kurachi H, Kim T, Enomoto T, Narumi Y, et al. MR imaging of mesenteric hemangioma: a case report. Radiat Med. 2000;18(1):67–69.
3. Hanatate F, Mizuno Y, Murakami T. Venous hemangioma of the mesoappendix. Surg Today. 1995;25(11):962–964. Available from: https://doi.org/10.1007/bf00312382.
4. Golitz LE. Heritable cutaneous disorders which affect the gastrointestinal tract. Med Clin North Am. 1980;64(5):829–846. Available from: https://doi.org/10.1016/s0025-7125(16)31569-3.
5. Schwartz GD, Barkin JS. Small bowel tumors. Gastrointest Endosc Clin N Am. 2006;16:267–275.
6. Lewis RJ, Ketcham AS. Maffucci's syndrome: functional and neoplastic significance. Case report and review of the literature. J Bone Joint Surg Am. 1973;55(7):1465–1479.
7. Arguedas MR, Shore G, Wilcox CM. Congenital vascular lesions of the gastrointestinal tract: blue rubber bleb nevus and Klippel-Trenaunay syndromes. South Med J. 2001;94(4):405–410.
8. Martínez JMA, Bellido CB, Macías MS, García-Moreno J, Grau JMS, Galindo AG. Massive mesenteric angiomatosis and low digestive hemorrhage in a

patient with Klippel-Trenaunay-Weber syndrome. Rev Esp Enferm Dig. 2007;99(2):112–113. Available from: https://doi.org/10.4321/s1130-01082007000200009.
9. Rodríguez-Castro KI, Antonello A, Ferrarese A. Spontaneous bleeding or thrombosis in cirrhosis: What should be feared the most? World J Hepatol. 2015;7(14):1818–1827. Available from: https://doi.org/10.4254/wjh.v7.i14.1818.
10. Levy AD, Abbott RM, Rohrmann CA, Frazier AA, Kende A. Gastrointestinal hemangiomas: imaging findings with pathologic correlation in pediatric and adult patient. AJR Am J Roentgenol. 2001;177(5):1073–1081. Available from: https://doi.org/10.2214/ajr.177.5.1771073.
11. Murphey M, Fairbairn K, Parman L, Baxter K, Parsa M, Smith W. From the archives of the AFIP. Musculoskeletal angiomatous lesions: radiologic-pathologic correlation. Radiographics. 1995;15(4):893–917. Available from: https://doi.org/10.1148/radiographics.15.4.7569134.
12. Costi R, Bian L, Smadja A, Violi C, V. A rare case of appendicitis-like syndrome: prompt laparoscopic diagnosis and management. J EmergMed. 2013;44(4):773–776.
13. Nader PR, Margolin F. Hemangioma causing gastrointestinal bleeding: case report and review of literature. Am J Dis Child. 1967;111:215–222.
14. Weinste EC, Moertel CF, Waush JM. Intussuscepting hemangiomas of the gastrointestinal tract: report of a case and review of literature. Ann Surg. 1963;157:265–270.
15. Ruiz AR, Ginsberg AL. Giant mesenteric hemangioma with small intestinal involvement: an unusual cause of recurrent gastrointestinal bleed and review of gastrointestinal hemangiomas. Dig Dis Sci. 1999;44(12):2545–2551. Available from: https://doi.org/10.1023/a:1026659710815.
16. Jun S, Tokihiro Y, Akihiro T. A case of giant diffuse cavernous hemangioma of the intestine and mesenterium associated with repeated kasabach-merritt syndrome. Journal of Japan Surg Assoc. 2002;63(3):723–772.
17. Oner Z, Altaca G. Diffuse cavernous rectal hemangioma-clinical appearance, diagnostic modalities and sphincter saving approach to therapy:report of 2 and a collective review of 79 cases. Acta Chir Belg. 1993;93(4):173–176.
18. Hanatate F, Mizuno Y, Murakami T. Venous hemangioma of the mesoappendix: report of a case and a brief review of the Japanese literature. Surg Today. 1995;25(11):962–964. Available from: https://doi.org/10.1007/bf00312382.
19. Amati AL, Hecker A, Schwandner T, Ghanem H, Holler J, Reichert M, et al. A hemangioma of the sigmoid colon mesentery presenting as a retroperitonealtumor: a case report and review. World J Surg Oncol . 2013;12:79. Available from: https://dx.doi.org/10.1186/1477-7819-12-79.
20. Chung J, Kim M, Jt L, Yoo HS. Cavernous hemangioma arising from the lesser omentum: MR findings. Abdom Imaging. 2000;25(5):542–544. Available from: https://doi.org/10.1007/s002610000087.

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