



MULTIPLE MESENTERIC LYMPHANGIOMAS: A CASE REPORT

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ABSTRACT Lymphangioma is a benign condition characterized by proliferation of the thin-walled lymphatic spaces. Incidence is 6% within the rest of the benign tumors. It is a congenital malformation. These are most frequently located in the cervical region 75%, followed by the axillary region in 20% of cases. 5% are located in abdominal region and within this the mesenteric location is the most common. Lymphangiomas are classified into three histologic types: capillary, cavernous and cystic. We report a case of a 2-year old boy who presented to our hospital in the surgery department with complaint of abdominal pain and vomiting. Whole Abdomen Ultrasonography revealed a well defined multicystic mass with few thin internal septae measuring 82x35mm with an impression of ?Lymphangioma ??Omental/mesenteric cyst. Mesenteric Lymphangioma is a rare tumor and multiple cysts are even rarer. Histopathology is the Gold standard in the diagnosis. With the best of our knowledge only few cases of multiple mesenteric lymphangiomas have been reported in the literature.

KEYWORDS :

INTRODUCTION:

Lymphangioma is a benign condition characterized by proliferation of the thin-walled lymphatic spaces [1]. Incidence is 6% within the rest of the benign tumors. It is a congenital malformation. These are most frequently located in the cervical region 75%, followed by the axillary region in 20% of cases [2]. 5% are located in abdominal region and within this the mesenteric location is the most common [3]. They occur mainly in pediatric patients in the first few years of life. It has abdominal complications such as bleeding, occlusive symptoms, recurrent abdominal pain and intestinal perforation [4].

Case presentation:

We report a case of a 2-year old boy who presented to our hospital in the surgery department with complaint of abdominal pain and vomiting for 15 days. These symptoms were progressively increasing. On examination there was abdominal distension with diffuse tenderness and hyperactive bowel sounds. The patient's vital signs were stable except for the pulse rate, 110 beats/min. Whole Abdomen ultrasonography revealed a well defined multicystic mass with few thin internal septae measuring 82x35mm is seen on right side of abdomen with an impression of ?Lymphangioma ??Omental/mesenteric cyst (less likely) was given. Laparotomy was performed. Surgical resection of part of small intestine along with mesenteric cyst was done. Milky white fluid was evacuated from one of the largest cyst. Specimen was sent in the 10% formalin to pathology department. On Gross examination (Figure A and B) received a segment of intestine measuring 8.5cm in length. On mesenteric border multiple cysts identified varying in diameter from 0.5cm to 2cm in diameter. On cutting open milky white fluid recovered. Cut section intestine-unremarkable. One lymph node identified measuring 0.5cm in diameter.



Figure A: showing thin wall multiple cysts of varying size at mesenteric border



Figure B: showing larger cut opened thin walled cyst at mesenteric border.

On microscopy (Figure C and D)- sections examined from the cyst show cyst wall comprising of fibromuscular tissue lined by flattened to cuboidal epithelium. Wall shows numerous dilated thin walled vessels filled with homogenous proteinaceous material and red blood cells. Mild inflammatory cell infiltrate comprising of lymphocytes and plasma cells is also noted in the wall. Random sections from small intestine show no significant pathology. Both resection limits were unremarkable. Lymph node shows Reactive Lymphoid Hyperplasia. Histopathological diagnosis was mesenteric lymphangioma.

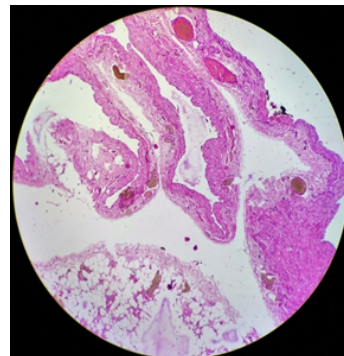


Figure C: cyst wall

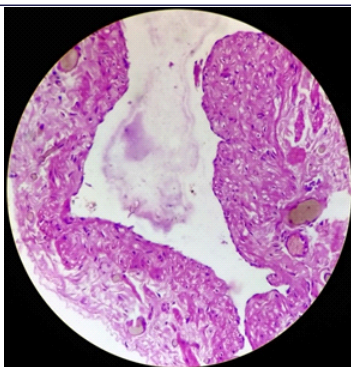


Figure D: cyst containing proteinaceous fluid

DISCUSSION

Lymphangioma is a rare benign tumor formed by vascular malformations due to failure of communication of lymph sacs with venous drainage system [5,6]. Predominantly head, neck and axillary regions are affected by the tumor. They can also appear rarely in the abdominal or mediastinal cavity, among this lymphangiomas of the small bowel mesentery has been described in <1% of lymphangiomas [6]. The clinical presentation of abdominal lymphangiomas are non-specific and may include abdominal pain, vomiting, abdominal distension, a palpable abdominal mass or acute abdomen. Mesenteric lymphangioma as a lymphatic vascular lesion has similar characteristics to extra-abdominal locations and its clinical behavior depends on its location. The theories of their congenital development has been accepted. The lymphatic system develops from the fifth week of the embryonic period with the formation of six lymphatic sacs and their further development depends on the intercommunication of these with the vessels and lymphatic capillaries in different areas of the body. The mesenteric location will most likely affect vasculature of the intestine. The locations in free organs as the omentum, the tumor will need a greater growth to produce compressive effect on the adjacent organs. Hence, the dimensions of the tumor location of the tumor and the age of the patient will be some factors that will condition the initiation of the clinical manifestations.

Lymphangiomas are classified into three histologic types: capillary, cavernous and cystic [6].

1. The Capillary type usually originates in the skin and consists of uniform small thin-walled lymphatic spaces.
2. The cavernous type is composed of various sizes of dilated lymphatic spaces associated with lymphoid stroma and shows a connection with the adjacent normal lymphatic spaces.
3. Cystic type consists of dilated lymphatic spaces of various sizes associated with collagen and smooth muscle bundles in the stroma but lacks connection to the adjacent normal lymphatic spaces [7].

Table 1: Comparison of Present study with other studies.

| Study | Age group | Clinical presentation | Gross Examination | Radiology |
|-------------------------------|----------------|--|--|---|
| Present Study | 2yrs | Abdominal pain, vomiting | Multiple cysts identified, larger measuring 2cm in diameter. | a well defined multicystic mass measuring 82x35mm is seen in right side of abdomen. |
| Rodríguez VC et al [8] | 12yrs and 4yrs | Intestinal obstruction | Cystic, multiloculated | Distended loop image in the upper abdomen |
| Rattan K N et al [9] | 18month -10yrs | Abdominal lump, pain, intestinal obstruction | Single, multiloculated | Multiloculated cystic lesions in relation to gut |
| Suthiwartnarueput W et al [5] | 2yrs, 9month | Abdominal distension and vomiting | Huge 20x20x10cm, lobulated and cystic | Signs of intestinal occlusion |

Table 1: shows that pediatric age group is the most commonly affected

by the mesenteric lymphangioma. A 2yrs old boy is affected by the tumor in our study which is in concordance to the studies of Rodríguez VC et al [8], K N Rattan et al [9], and Suthiwartnarueput W et al [5] mesenteric lymphangioma. Clinical manifestations are abdominal pain and vomiting that is almost similar to the findings of Rodríguez VC et al [8], Rattan K Net al [9] and Suthiwartnarueput W et al [5]. On gross examination Rodríguez VC et al [8], Rattan K N [9] and Suthiwartnarueput W et al [5] found single mass/cyst. Here our study differs, there were multiple cysts involving the small bowel mesentery varying in size from 0.5cm to 2cm. Radiology shows a multicystic mass in present study as well as in studies conducted by the Rodríguez VC et al [8], Rattan K N [9] and Suthiwartnarueput W et al [5].

Diagnosis is practically impossible on clinical grounds [5, 6]. The development of imaging techniques to date has allowed the diagnosis of these cystic tumor lesions from the fetal stage. The use of simple abdominal radiographs will be more associated with the diagnosis of the abdominal pain in its acute phase, being able to find signs of intestinal occlusion and radiopacity of an abdominal area with changes in the distribution of the gas pattern. Also intra-abdominal calcifications could be defined with this medium. Histopathology is the gold standard for the diagnosis of various types of lymphangioma. The treatment of abdominal lymphangiomas is surgical excision. Incomplete resection usually leads to recurrence in ~10–15% [10].

CONCLUSION:

1. Mesenteric Lymphangioma is a rare tumor and multiple cysts are even rarer.
2. Histopathology is the Gold standard in the diagnosis.

With the best of our knowledge only few cases of multiple mesenteric lymphangiomas have been reported in the literature.

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