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# Cystic Lymphangioma of Mesocolon: A Rare Site and Fate

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**Abstract** **Case Report**

Lymphatic malformations (LM) are rare benign tumours that falls under the larger umbrella of vascular tumours. Almost 90% of the cases are diagnosed within the first two years of life. Here we present two year old boy who presented acutely with abdominal pain and fever with tenderness over the right lower quadrant. Ultrasonography and computed tomography of the abdomen revealed a large intra-peritoneal multicystic lesion. Patient underwent an elective laparotomy and excision of the lesion which was situated at left mesocolon. Intra-operative measurement of the tumour was only one third the size on initial imaging, which indicates a partially regressed tumour. Histopathological examination confirmed the diagnosis of cystic lymphangioma (CL). Patient was well post-operatively and discharged within 5 days. We report a patient with CL at a rare site with partial regression managed successfully with surgical excision.

**Keyword:** Lymphatic malformations (LM), vascular tumours, Ultrasonography, cystic lymphangioma (CL).



## 1. INTRODUCTION

Lymphatic malformations are benign low flow vascular variances of the lymphatic system. The incidence is found to occur in one out of 2000 to 4000 live births [1]. 50 to 60% of these lesions are diagnosed by the first year of life, and 80 to 90% are diagnosed by the age of two. Advances in antenatal screenings and developments in imaging techniques and skills have made it possible for accurate prenatal identification of these lesions. Cystic lymphangioma are uncommon tumours, and may be found at various anatomical regions. Three quarter of CL are found at head and neck due to their rich lymphatic channels, 20 percent in the axillary region and five percent in rest of the areas, making abdomen an unusual site [3]. This makes intra-abdominal CL's incidence between 1 in 20,000 to 250,000. Intra-peritoneal CLs are usually found at mesentery of small bowels, followed by omentum and rarely in the mesocolon [4]. They are mainly classified as microcystic, macrocystic or combined macro- and microcystic lesions depending on size with 1cm being the cut-off [5]. They commonly present as palpable mass with a number of them having pressure effect on adjacent organs. Various treatment modalities can be used in the management of these tumours. We herein report a case of CL found in an unusual site with partial regression requiring surgical excision.

## 2. CASE REPORT

A 2 years old boy presented with history of low grade fever and lower abdominal pain for four days. There was no other significant positive history. He was still able to ambulate and did not show any sign of dehydration. His pain score was 4 according to the Wong Baker facies scale. Palpation of his right lower quadrant of the abdomen was tender with no signs of peritonism. His full blood count showed leucocytosis of 21,000/L. At this time, a diagnosis of possible appendicitis was made. An ultrasonography (USG) of the abdomen revealed a large heterogenous multicystic lesion with predominant hyperechoeic components occupying the lower quadrant of the abdomen (Figure 1). Subsequently, a contrast enhanced computed tomography (CT) of the abdomen was performed. There was a large, well defined multiloculated intra peritoneal cystic lesion measuring 8cm x 7cm x 13cm occupying left lower abdomen (Figure 2). He was then managed conservatively as his symptoms improved and parents were not keen for any surgical intervention. An outpatient assessment a month later revealed persistent intermittent abdominal pain, thus he underwent laparotomy and tumor excision. Intra-operatively, there was a multicystic lesion measuring 5cm x 2.5cm x 2.5cm at the left mesocolon with multiple mesenteric lymph nodes (Figure 3). These lesions were then sent for histopathological examination (HPE) and confirmed a cystic lymphangioma. Lymphoid aggregate,

multinucleated giant cells, macrophages and lymphocytes were present. There was no evidence of necrosis, cellular atypia or malignancy. He recovered well post operatively and was discharged on day five. He was followed up a month later and did not have any abdominal complaints.

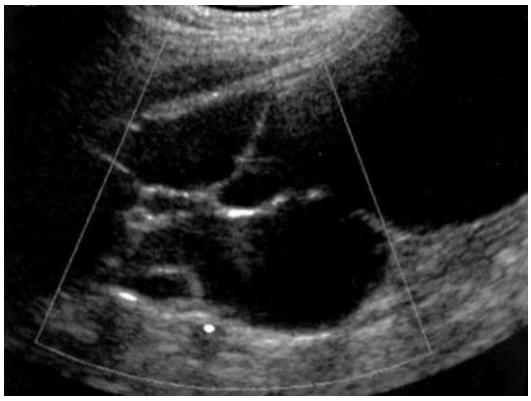

**Figure 1: Ultrasonography of the abdomen showing a large heterogenous multicystic lesion occupying the lower quadrant**

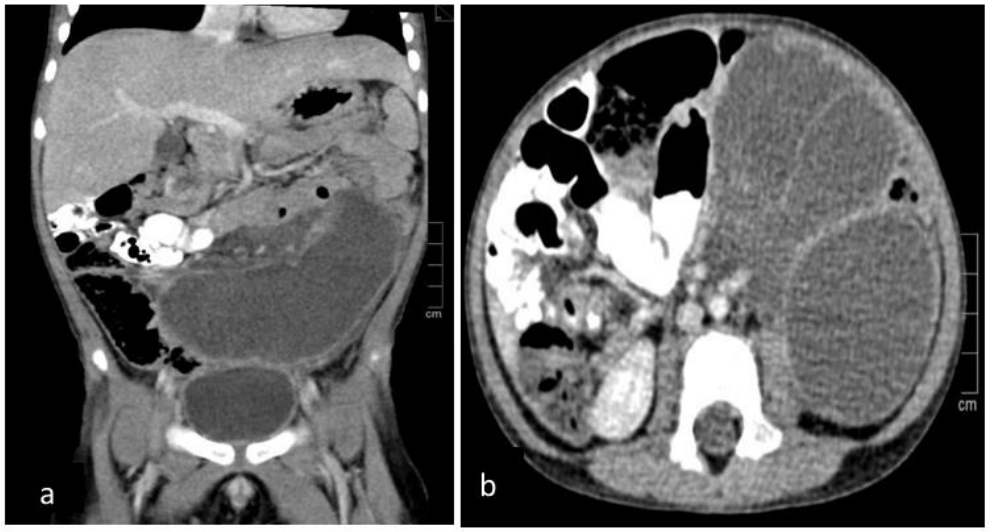


**Figure 2 (a-b): Contrast enhanced computed tomography (CT) of the abdomen in coronal and axial views showing a well-defined multiloculated intra peritoneal cystic lesion measuring 8cm x 7cm x 13cm occupying left lower abdomen**

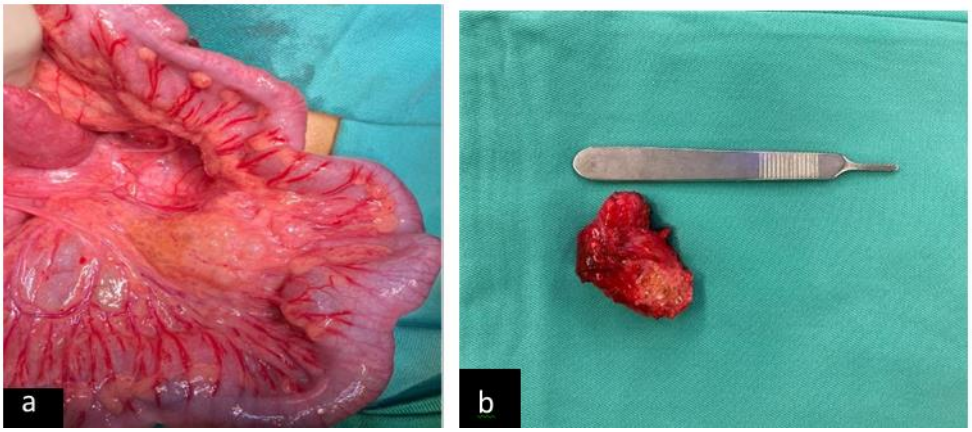


**Figure 3 (a-b): Laparotomy findings of a cystic tumour at the left mesocolon measuring 5cm x 2.5cm**

## 3. DISCUSSION

Cystic lymphangiomas are benign lesions of the lymphatic system and falls under the larger umbrella of vascular anomalies. Previously, various inconsistent nomenclatures were used to identify this rare tumour, which includes cystic hygroma, lymphangioma, lymphangiomatosis or even hemangioma [6]. International Society for the Study of Vascular Anomalies (ISSVA) sub- classified CL according to the sizes of cystic spaces into microcystic, macrocystic or mixed in 2014. The aetiopathogenesis is not fully

understood, however a few postulations have been made. These includes failure of embryonic lymphatic system to communicate with venous system, genetic abnormalities of lymphatic cells and parts of sequestered sacs from primary lymphatic channels during embryonal development. Acquired CL arises from lymphatic obstruction due to triggering factors like trauma, infection and radiotherapy.

Advent of technology has increased the prenatal diagnosis of these tumours. Most children with abdominal CL are usually asymptomatic and left undiagnosed unless complicated with an acute complication. These includes abdominal discomfort, abdominal distension, fever, intestinal obstruction or peritonitis [7]. These tumours can also reveal after a viral or bacterial infection. Concurrent cutaneous manifestations such as lymphatic papules and capillary malformations have also been reported. Large tumours commonly present as a ballotable mass.

A number of imaging modalities can be used to diagnose these lesions, mainly USG and CT. USG demonstrates thin walled, multi-septated structures without peristalsis, and Doppler US demonstrates no blood flow within the cystic walls. CT is crucial when diagnosis is in doubt and the need to rule out other sinister differentials such as teratomas.

Treatment strategies for intra-abdominal CL evolve mainly around surgical excision and sclerotherapy, with the former being the treatment of choice. Recurrences are reported as 12% and 53% respectively for complete and partial resection. As it is a benign condition, benefit of aggressive excision needs to be well-adjusted to associated risk of injuring adjacent structures or bowel. On the other hand, sclerotherapy is reserved for unresectable tumours or those confined to head and neck. Spontaneous regression has also been reported in certain cases and needs to be considered on a case by case basis. A study concluded that macrocystic and mixed type CL has possibility of spontaneous regression and suggested to wait for treatment for more than three months, anticipating a regression, while microcytic should receive prompt treatment due to low likelihood of spontaneous regression [8].

Peculiarity of this case is that the child came with symptoms of what appears to be an acute abdomen. Ultrasonography of the abdomen however revealed a large cystic lesion occupying the lower abdomen necessitating a computed tomography, in suspicion of a malignancy. The initial dimensions were 8cm x 7cm x 13cm, v during surgery the tumour has shrunk to almost one third of the initial size. This partial regression too two months. Initial leucocytosis, pain and fever points towards a bacterial superinfection of an existing congenital CL. Secondly, the child developed the tumour along the left mesocolon which is regarded as the least common site of occurrence among those intra-peritoneal CL compared to small bowel mesentery and omentum.

## CONCLUSION

Lymphatic malformations are rare and can be a challenging diagnosis to clinicians as presenting symptoms can be non -specific. Diagnosis needs to be made with an imaging modality and direction of treatment must be taken considering the size of the tumour and manifestations as some of these CLs can be managed non- operatively with regular imaging follow up. Surgical resection is still considered the best option when dealing with resectable symptomatic tumours.

**Disclosure:** The authors have nothing to disclose.

## REFERENCES


1. Kennedy, T. L., Whitaker, M., Pellitteri, P., & Wood, W. E. (2001). Cystic hygroma/lymphangioma: a rational approach to management. *The laryngoscope*, *111*(11), 1929-1937.
2. Stessy, K., Diana, P., Khalil, C., Emmanuelle, V., Marie, A., Guillaume, L., & Jiad, N, M. (2020). Retroperitoneal Cystic Lymphangioma: A Case Report of Spontaneous Regression and Review of Literature. *Dermatology Archives*, *4*(1).
3. Chew, B. J. W., Dagash, H., & Khare, M. M. (2019). Intra-abdominal cystic lymphangioma. *The Journal of Pediatrics*, *205*, 288-288.
4. Kati, Ö., Güngör, Ş., & Kandur, Y. (2018). Mesenteric cystic lymphangioma: Case report. *Journal of Pediatric Surgery Case Reports*, *35*, 26–28.
5. Bagrodia, N., Defnet, A. M., & Kandel, J. J. (2015). Management of lymphatic malformations in children. *Current Opinion in Pediatrics*, *27*(3), 356–363.
6. Wohlgemuth, W. A., Brill, R., Dendl, L. M., Stangl, F., Stoevesandt, D., & Schreyer, A. G. (2018). Abdominal lymphatic malformations. *Der Radiologe*, *58*(S1), 29–33.
7. Yokoyama, S., Sekioka, A., & Utsunomiya, H. (2014). Spontaneous regression of a cystic peripancreatic tumor in a 12-year-old boy: A case report. *Journal of Pediatric Surgery Case Reports*, *2*(6), 297–301.
8. Kato, M., Watanabe, S., Kato, R., Kawashima, H., Iida, T., & Watanabe, A. (2017). Spontaneous Regression of Lymphangiomas in a Single Center Over 34 Years. *Plastic and Reconstructive Surgery - Global Open*, *5*(9).