ORIGINAL RESEARCH PAPER
Volume-9 | Issue-5 | May-2020 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

A RARE CASE OF CYSTIC LYMPHANGIOMA IN AN ADULT FEMALE

General Surgery

Dr. G. Muthukumaran	M.S
Dr. M. Shobana*	*Corresponding Author
Dr. Touzeen Hussain	M.S
Dr. Sandeep Naidu	
Dr. Sivaram	
Dr. V.S. Bubeshver	
Dr. Sudarshan S Murthy	

KEYWORDS

INTRODUCTION

Cystic lymphangiomas, mostly occurring in children, are rare lymphatic developmental anomalies. Most frequent location is the head and neck, while intra-abdominal location is an extremely rare entity. Cystic Lymphangioma is an uncommon, slowly growing tumour derived from the lymphatic vessels. Although the aetiology remains unclear, lymphangiomas are thought to result from a developmental failure of the lymphatic system. They are often confused with mesenteric cysts that arise from mesothelial, not lymphatic tissue. We report a case of cystic lymphangioma of the lesser sac in a 43-year-old woman.

CASE REPORT

A 43 year-old woman presented with a 1 year history of mild epigastric, dragging type of intermittent pain, not associated with food intake. No history of loss of weight, loss of appetite. No history of nausea, vomiting, altered bowel or bladder habits. On examination, mild epigastric tenderness was noted with no other physical signs. Abdominal ultrasound revealed a multiseptated focal cystic lesion in the gastrohepatic region, measuring 43 x 55 x 42mm. An abdominal contrast enhanced computed tomography showed an intra peritoneal, focal non-enhancing, cystic lesion/area seen in the gastrohepatic region (lesser omentum) of size ~ 48 x 58 x 44mm, volume- 61cc, central variable density (-4 to 30HU), eccentric focal tiny calcification, encasing left gastric artery (no filling defect) peripheral lobulated to irregular margins and abutting lesser curvature of stomach, caudate and left lobe of liver (no invasion). Routine laboratory investigations were within normal limits.

Laparoscopic exploration followed by laparotomy with complete excision of the cystic lesion located in the lesser omentum with an intimate relation to the left gastric artery, containing clear fluid was done.

Cut surface of the specimen showed grey brown to yellowish with tiny cystic areas, largest measuring 1cm, smallest measuring 0.2cm. Histopathologic examination showed fibrofatty tissue and multiple dilated lymphatic vessels filled with lymphocytes, congested vessels and hemorrhagic fluid. Features suggestive of a cystic lymphangioma.

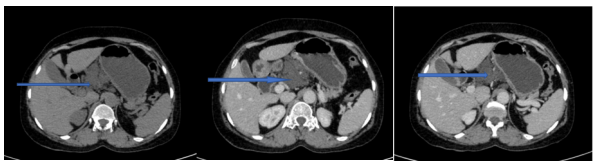

Figure 1 CECT of the Abdomen, Axial cuts showing an intra-peritoneal, non-enhancing, focal cystic lesion, in the gastrohepatic region with eccentric focal tiny calcification.

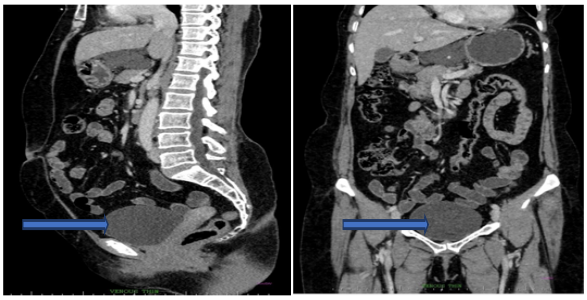

Figure 2 CECT of the Abdomen, Coronal and Sagittal sections showing an ill defined cystic lesion encasing the left gastric artery and abutting the lesser curvature of stomach, caudate lobe and the left lobe of the liver.

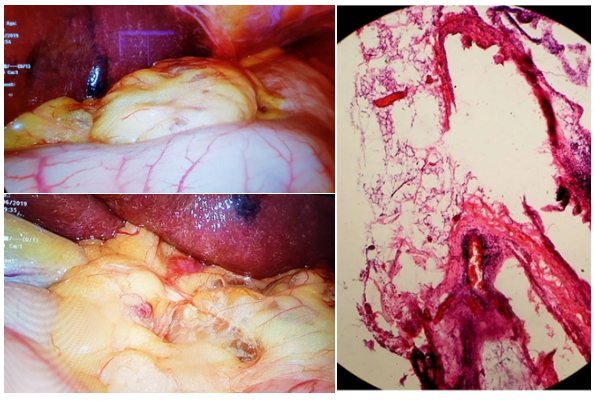

Figure 3. Intra-operative images

fibrofatty tissue and multiple dilated lymphatic vessels filled with lymphocytes, congested vessels and hemorrhagic fluid. Features suggestive of a cystic lymphangioma.

DISCUSSION

Intra-abdominal cystic lymphangiomas are not common. Abdominal cystic lymphangiomas are rare tumours with a reported incidence between 1 in 20,000-250,000 (4,6). Almost 90% are detected by the mean age of 2 years, and most occur in the small bowel mesentery (4,7). Their location can involve the mesentery, omentum, colon, spleen, pelvis, groin and retroperitoneum (1,8). Cystic lymphangiomas are considered to originate from malformed or malpositioned lymphatic tissue, but other factors such as abdominal trauma, lymphatic obstruction, inflammatory process, surgery, or radiation therapy may lead to secondary formation of such tumours

(1,9). Large series of intra-abdominal cystic lymphangiomas are rarely reported in the medical literature. Less than 1% of the cystic lymphangiomas are found in the retroperitoneum, and the rare lymphangiomas occurring in the mesentery have been classified into six groups by de Parrot et al (2,10). Losanoff et al (2,11). Defined four different types of mesenteric cystic lymphangiomas, including pedicled, sessile, retroperitoneal extended, and multi-centric lymphangiomas. Acute abdomen is seldom reported in adults (2,11). The clinical presentation is variable. Asymptomatic course is usual, but it can also be the cause of non-specific symptoms, such as anorexia, nausea, vomiting, fatigue, and weight loss. Chronic abdominal distension, detected by palpation of a cystic mass may be present. Acute abdominal pain requiring surgery is not common (1,9,12).

The aetiology of mesenteric lymphangioma is considered to be congenital, with abnormal embryonic development of the lymphatic system causing sequestration of lymphatic tissue (3,13). Histopathologically, abdominal cystic lymphangiomas are characterised by a thin irregular wall-covered by endothelium that contains smooth muscle, foam cells, and lymphatic tissue (4,7,14). They are often confused with mesenteric cysts that arise from mesothelial, not lymphatic, tissue. This differentiation is important because lymphangiomas often behave in an invasive and aggressive manner, whereas mesothelial cysts do not. Despite being difficult to differentiate on imaging studies, they are histologically distinct from one another. Lymphangiomas have an endothelial lining, foam cells, and a wall that contains lymphatic spaces, lymphoid tissue, and smooth muscle. Mesenteric cysts either have no lining or are lined with cuboidal or columnar epithelium. (6) However, other possible causes have been suggested such as: abdominal trauma, lymphatic obstruction, inflammatory process, surgery, and/or radiation therapy (3,13). The differential diagnosis includes cystic lesions of mesothelial, enteric or urogenital origin, Dermoid cysts or teratomas, pseudocysts from trauma or infectious origins, and cystic degeneration of solid tumours (1,10,15,16).

MRI and CT are common diagnostic modalities, and the typical diagnostic criteria were cystic mass and presence of septa (2,16). The ultrasonographic presentation of a mesenteric lymphangioma is described as a well-circumscribed cystic anechoic lesion with irregular thin walls and multiple thin septa (honeycomb or cobweb pattern)(4,5). On CT imaging, mesenteric lymphangiomas appear as a uni- or multiple locular masses with enhancement of the wall and septum by contrast medium (3,17).

The definitive diagnosis is always made histologically (1,3,4). Lymphangiomas do not generally regress (1,18). The main modality of treatment of mesenteric lymphangioma is adequate surgical excision, because there is a risk of recurrence and malignant transformation, particularly after radiotherapy for the primary lesion (4,11). Complete surgical excision is the treatment of choice for cystic lymphangioma, even if asymptomatic (3). Depending on the size and location of the mass, laparoscopic resection has been proposed as a safe and minimally invasive means of surgical treatment.

Complete removal of the tumour offers an excellent prognosis (1,3). On the other hand, recurrence has been reported in 10% of patients in whom primary resection was incomplete. If the stem of the cyst and feeding lymphatics are unsuccessfully ligated, chylous ascites may also occur. Isolation and ligation of lymph channels can prevent recurrences and chylous ascites. Recurrence may necessitate a second operation (1,19).

CONCLUSION

In conclusion, intra- abdominal cystic lymphangiomas are rare entities. Preoperative diagnostic tools are Ultrasound and CT or MRI of the abdomen. Early recognition and appropriate treatment of these cysts are associated with a good prognosis. Complete surgical resection is the ideal modality of treatment for mesenteric lymphangioma. Definitive diagnosis is confirmed by histopathology. However, long term follow-up is advisable because of the possibility of recurrence.

REFERENCES

1. C Papanicolaou, G Anthimidis, DAlataki, V Lagopoulos, I Sougas, G Vaios, G Hatzitheoharis. Cystic lymphangioma of the lesser sac (omental bursa). The Internet Journal of Surgery. 2008 Volume 18 Number 1.
2. Chih-Ming Su, Ming-Chin Yu, Huang-Yang Chen, Jeng-Hwei Tseng, Yi-Yin Jan, Miin-Fu Chen. Single-Centre Results of Treatment of Retroperitoneal and Mesenteric Cystic Lymphangiomas.
3. Giovanni Aprea, Francesco Guida, Alfonso Canfora, Antonio Ferronetti, Antonio Giugliano, Melania Battaglini Ciciriello, Antonio Savanelli, Bruno Amato. Mesenteric Cystic Lymphangioma in adult: A case series and review of the literature.
4. Omer Kati, Sukru Gungor, Yasir Kandur. Mesenteric Cystic Lymphangioma: Case Report.
5. F. Geoff Allen, M>D., Taylor Sohn Riall, M.S., Fohn L. Cameron, M.D., Frederic B. Askin, M.D., Ralph H. Hruban, M.D., Kurt A. Campbell, M.D. Abdominal Lymphangiomas in Adults.
6. Kurtz RJ, Heimann TM, Holt J, Beck AR. Mesenteric and retroperitoneal cysts. Ann Surg 1986;203:109-12.
7. Gosh BK, Tan YM, Ong HS, Chui CH, Ooi LL, Chow PK, Tan CE, Wong WK. Intra-abdominal and retroperitoneal lymphangiomas in paediatric and adult patients. World J Surg 2005;29:837-40.
8. Fisher D, Hiller N. Case report: giant tuberculous cystic lymphangioma of posterior mediastinum, retroperitoneum and groin. Clinical Radiology 1994; 49: 215-6.
9. Daniel S, Lazarevic B, Attila A. Lymphangioma of the mesentery of the jejune: report of a case and a brief review of the literature. Am J Gastroenterol. 1983; 78: 726-9.
10. De Parrot M, Brundler M, Tostch M, Mentha G, Morel P: Mesenteric cysts. Toward less confusion? Dig Surg 2000;17:323-328.
11. Losanoff JE, Richman BW, El- Sherif A, Rider KD, Jones JW: Mesenteric cystic lymphangioma. J AM Coll Surg 2003;196:598-603.
12. Steyaert H, Guitard J, Moscovici J, Juricic M, Vaysse P, Juskiewenski S. Abdominal cystic lymphangioma in children: benign lesions that can have a proliferative course. J Pediatr Surg 1996; 31: 677-680.
13. Roisman I, Manny J, Fields S, Shillong E: Intraabdominal lymphangiomas. Br J Surg 1989, 76:485-489.
14. Alqahtani A, Nguyen LT, Flageole H, Shaw K, Laberge JM. 25years' experience with lymphangiomas in children. J Pediatr Surg 1999;34:1164-8.
15. Stoupis C, Ros PR, Abbott PL, Burton SS, Gauger J. Bubbles in the belly: imaging of cystic mesenteric or omental masses. Radio graphics. 1994; 14: 729-37.
16. Yang DM, Jung DH, Kim H, Kant JH, Kim SH, Kim JH, Hwang HY. Retroperitoneal cystic masses: CT, Clinical, and Pathological Findings and Literature Review. RadioGraphics 2004;24: 1353-1365.
17. Davidson AJ, Hartman DS: Lymphangioma of the retroperitoneum. CT and sonographic characteristics. Radiology 1990, 175:507-510.
18. Duthler RJ, Fisichella PM. Image of the month. Cystic lymphangioma. Arch Surg. 2005; 140: 705-6.
19. Okur H, Kucukaydin M, Ozokutan BH, Duran AC, Kameez A, Lose O. Mesenteric, omental, and retroperitoneal cysts in children. Eur J Surg 1997;163: 673-677.