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CASE REPORT

Mesenteric Cystic Lymphangioma in an Adult Female - A Case Report

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ABSTRACT

Introduction: Lymphangiomas are benign vascular proliferations composed of malformed lymphatics, located in the head, neck, and axilla. Incidence in adults and at the retroperitoneal location is rare.

Case Presentation: We present a 34-year woman with a 7-day history of abdominal distention, colicky pain, and vomiting. Physical examination shows a non-tender, diffusely enlarged abdomen and absent bowel sounds. She underwent a magnetic resonance imaging (MRI), which confirmed 18.0 x 14.3 x 8.5 cm, multi-loculated mass extending along the mesentery of the small intestine. On histopathological examination cystic lymphangioma was the final diagnosis. Cystic lymphangioma is an unusual lesions associated with genetic and chromosomal abnormalities. Radiological investigations play a key role in pre-operative diagnosis. Final diagnosis can be obtained only after histopathological evaluation.

Conclusion: This report adds to current knowledge on this pathology in atypical sites by providing thorough insights into clinical aspects, MRI results, and the histopathological correlation.

Keywords: Adult, histopathology, mesenteric lymphangioma.

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Introduction

Lymphangiomas are benign localized or diffuse vascular proliferations composed of lymphatic vessels. They are preferentially located in the head, neck, and axilla in children, and in adults, and at the retroperitoneal location is rare (1).

However, lymphangiomas in the abdomen are extremely rare, particularly in adults. In the abdomen, lymphangiomas occur in the mesentery, followed by the omentum, mesocolon, and retro peritoneum. The etiology is unclear, but they are considered primarily to be congenital in origin. (2) Preoperative diagnosis is often difficult due to the frequent silent clinical course. Radiological investigations are a useful diagnostic tool, but definitive diagnosis is confirmed by histopathology after a complete surgical resection.(3) This report describes a case of an adult patient with mesenteric lymphangioma.

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Case Presentation

We report a 34-year-old female who presented with a 7-day history of abdominal distention, colicky pain, and vomiting. Past medical history was significant with recurrent

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complaints of nausea, bloating, and gradual abdominal distention. No previous trauma or surgery documented. Physical examination reveals a non-tender and enlarged abdomen. On palpation, a vague mass was noted deep in the abdomen. Blood, complete picture, tumor markers, and radiological studies were done. Her CRP, CA 125, CA 19-9, serum amylase, and CEA were in the normal range. Ultrasound reveals a 10cm mass behind the intestine. She underwent a magnetic resonance imaging (MRI), which confirmed the 10.0 x 8.3 x 5.5 cm, multi-loculated mass extending along the mesentery of the small intestine. Apparently, it was not originating from the stomach, pancreas, intestine, or ovaries. The loops of intestine were displaced and partially obstructed. Preliminary diagnosis of cystic lymphangioma was made.

The mass was excised, and the patient was discharged in stable condition after one week. Mass was submitted for histopathologic examination. Grossly, it was lobulated and reddish brown. Cut surface shows multiple locules filled with milky thick fluid. Microscopy reveals variable-sized spaces lined by flat endothelial cells. Stroma was fibrotic and foci of lymphoid aggregates with occasional collections of macrophages. On immunohistochemistry, endothelial cells were positive for CD 31, D2-40, and negative for Pan cytokeratin, WT 1, CK5/6 & Calretinin. The tumor was labelled as a cystic lymphangioma.

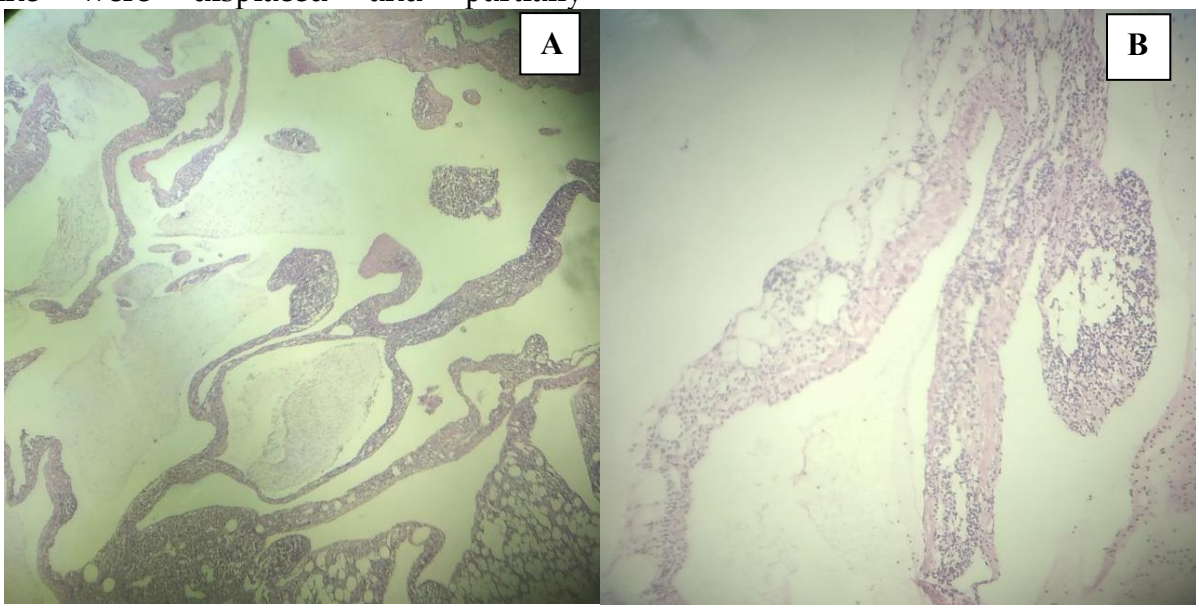

Figure-1A: Histopathological image showing the characteristic architecture of lymphangioma (Magnification: 4×). 1B. Large vascular spaces lined with endothelial cells containing eosinophilic fluid and mature lymphocytes and lymphoid aggregates. Staining: hematoxylin and eosin stain (Magnification: 10×).

Discussion

Hygroma in Greek refers to a water-filled tumour. Originating from improper separation of lymphatic tissue from sacs during development (3). Genetically, many cases show chromosomal abnormalities, including trisomy 21, trisomy 18, trisomy 13, Turner syndrome or Noonan syndrome. More recently, somatic mutations in PIK3CA have been identified([4) It represents anomalous vascular proliferation with a prevalence of 1.2 - 2.8 cases per 100,000 children. In adults, external causes such as radiation therapy, surgery, infection, or local trauma may contribute to the pathogenesis (5).

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Historically, the neck is the most frequent site (75%), but it can also be found in the axilla (20%) and less often in the mediastinum, retroperitoneum, mesentery, omentum, colon, pelvis, scrotum, and spleen. Retroperitoneal emplacement is rare and accounts for only 1% of lymphangioma (6). Here, the tumor may cause diagnostic dilemmas with other retroperitoneal cystic tumors, including those arising from the liver, kidney, pancreas, or ovaries (7). Rowe SP recently highlighted the adrenal gland as another rare location in his 2015 study.

Although benign and found accidentally during routine workup, they can grow to be quite large and cause symptoms as a result of their compressive effects on surrounding organs. Clinical presentation varies with the location of the tumor. (8) When the lesion is placed in the abdomen, common symptoms include vomiting, dyspepsia, abdominal discomfort, and intestinal blockage leading to abdominal distension. (9)

Pathologically classified into three basic types: cystic, capillary, and cavernous, however, other identified patterns include micro cystic, macro cystic, or mixed. (10) In suspected cases, ultrasonography should be performed first, followed by contrast-enhanced MRI. On CT scan, these lesions remain ambiguous, especially at odd locations for most radiologists. (8) Thus, MRI is regarded as the most effective non-invasive diagnostic modality. Most of the cases exhibit a lack of internal amplification and general uniformity, indicating their benign nature. (11)

Clinically, the differential diagnosis varies depending on the site of the lesion, which may include cystic lesions emerging from adjacent organs. Pseudocyst, epithelial cyst, parasite cyst, intussusception, haematoma, and dermoid cyst can all be found in the retro-peritoneum. (11) Mesothelioma, another key issue, appears as fluid-filled locules that resemble lymphangiomas on ultrasound and MRI. However, the principal differential diagnoses on clinico-radiological grounds are cystic lymphangioma and cavernous hemangioma, which can only be ruled out by histopathologic examination or EUS-guided aspiration in deeper locations. (12)

Cystic hygromas can be left untreated if it is of small size and causes no compressive effects. If treatment becomes necessary, then besides surgical intervention, sclerotherapy, radio ablation, laser excision, or cauterization may be considered where practicable. (13)

Conclusion

Although lymphangioma is rare, it should be considered as a differential diagnosis for any mass. Though most cases appear before the age of two, in the cervicofacial region, atypical locations and delayed presentation can occur. This research adds to current knowledge of this pathology in atypical sites by providing thorough insights into clinical aspects, MRI results, and the histopathological technique used.

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Ethical Approval

Approval was granted by the ethics committee of Watim Medical and Dental College.

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All the authors agree to take responsibility for every facet of the work, making sure that any concerns about its integrity or veracity are thoroughly examined and addressed.	