CASE REPORT

Mesenteric cyst of Mullerian origin: A case report and review of literature

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Abstract

Mesenteric cysts are rare entities, and the Mullerian origin subtypes even rarer. They are classified on the basis of their origin and morphology. The gold standard test for diagnosis is histopathology, whereas radiological imaging provides only supportive findings. The treatment of choice is surgical excision. We present the case of a 42-year-old female who presented in the surgical emergency of Mansoorah Teaching Hospital, Lahore, on August 16, 2022, with acute abdominal pain associated with nausea and vomiting. She was initially diagnosed as a case of simple unilocular left-sided ovarian cyst based on clinical symptoms and radiological findings. However, biopsy revealed a mesenteric cyst of Mullerian origin. As mesenteric cysts are not common, therefore they becomes extremely challenging for the radiologist, pathologist, and surgeon equally, due to the difficulties encountered in their diagnosis and management. We, hereby, present the much-needed literature review of these cysts with special emphasis on reproducible classification of mesenteric cysts and their clinical implications.

Keywords: Mesenteric cyst, Mullerian, Diagnosis, Histopathology, Surgery.

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Introduction

Cysts arising from mesentery, omentum, and mesocolon are known as mesenteric cysts.[1] Mesenteric cysts, first described at autopsy by Benevenni in 1507[1,2] are rare entities and the reported prevalence in the adult population varies from 1 per 100,000 to 250,000 admissions.[3]

The mesenteric cysts are classified based on the aetiology and morphological features.[2] The presenting complaints are usually non-specific and the radiological imaging results also fail to show any specific diagnostic features. In most cases, the diagnosis is confirmed after surgical excision and histopathology of the cyst. The proposed theories regarding the aetiology of mesenteric cysts are: (1) misplaced lymphoid tissue, (2) benign proliferation of ectopic lymphatics with limited or no drainage, (3) mechanical/anatomical factors causing obstruction of lymphatics, (4) traumatised lymphatics with subsequent uncontrolled growth, (5) improper or failed mesenteric leaves fusion, (6) degeneration of lymph nodes, and /or (7) intestinal diverticula which can transform into mesenteric cyst.[4] The treatment of choice is always surgical excision.[1]

The case of patient with an adnexal cyst finally diagnosed as a mesenteric cyst of Mullerian origin on the basis of the clinical history, histopathological and immunohisto-chemical findings is presented.

Case report

A 42-year-old female presented in the Emergency Department of Mansoorah Teaching Hospital, Lahore, on August 16, 2022, with acute abdominal pain associated with persistent nausea and anorexia for the last 6-8 weeks. She had no history of fever, vomiting or recent abdominal trauma and her menstrual cycles were unremarkable. Her abdominal examination revealed swelling and tenderness in the upper left quadrant. Ultrasound of the pelvis was ordered which showed a uterine fibroid and a large oval cystic anechoic space with no internal septations or soft tissue component near the left adnexa, concluding it as a unilocular left adnexal cyst. A provisional diagnosis of a simple unilocular left-sided adnexal cyst was made based on the ultrasound findings, which also showed uterine fibroids. Exploratory laparotomy was done and a cystic lesion adherent to the gut was observed in the left adnexal region, intraoperatively. The cyst was successfully excised and separated from the gut and the sample was sent for histopathology. The patient's condition remained stable during her four-day hospital stay following the surgery. She was then discharged in good medical condition with a cover of oral antibiotics and analgesics till the follow-up at two weeks. The sample was sent to the Department of Histopathology, University of Health Sciences, Lahore, for histopathological examination which revealed a cyst measuring 5.5cmx5.0cmx3.5cm with adherent left adnexa on gross inspection. (Figure 1). The cyst was unilocular and thin walled, containing greenish brown fluid. Microscopic examination showed a cyst lined by cuboidal to low

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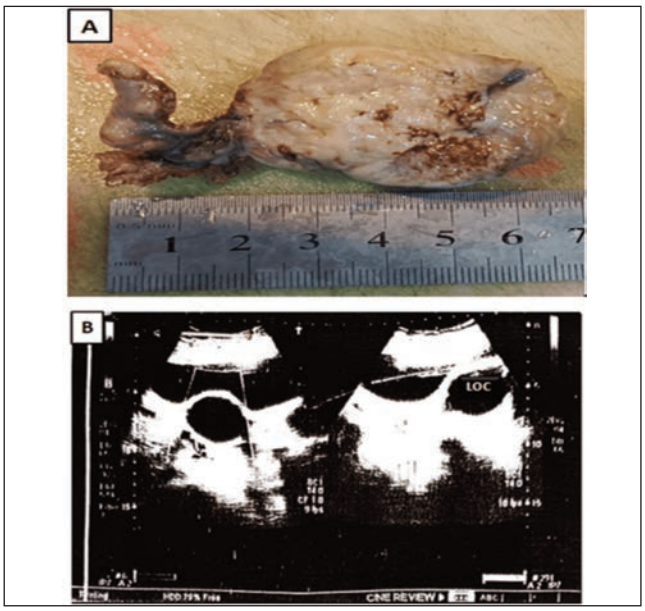

Figure-1: Image A shows the resected specimen of the left adnexal mass. Image B shows the ultrasound of left ovarian cyst which reveals a unilocular large oval cystic anechoic space with no internal septations or soft tissue component.

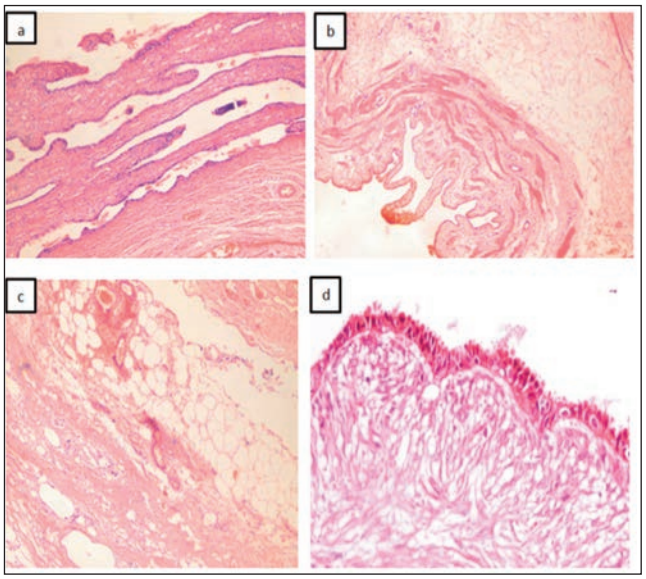

Figure-2: Photomicrograph of H & E stained sections (20X) of mesenteric cyst wall showing (a) cyst lined by ciliated cuboidal to low columnar epithelium at places forming tiny papillary tufts (b) smooth muscle bundles and fibro-adipose tissue (c) fibro-vascular and adipose tissue (d) The high resolution (40X) of the cyst lining reveals ciliated cuboidal to low columnar epithelium.

columnar ciliated epithelium. The underlying fibro-vascular wall had few smooth muscle fibres and lymphatic channels. In addition, mild chronic inflammatory cell infiltrate was also seen. Adherent ovary and tube were unremarkable (Figure 2 a-d). On immunohistochemistry, the lining epithelium of the cyst gave strong nuclear positivity for oestrogen and progesterone (ER/PR) hormone receptors (Figure 3 a & b). This case was diagnosed as mesenteric cyst

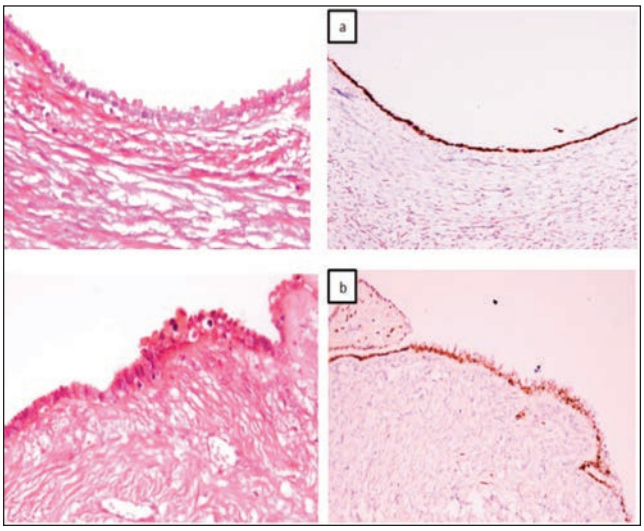

Figure-3: Photomicrograph of H & E stained sections (40X) of mesenteric cyst wall showing ciliated low cuboidal to columnar epithelium. Figure 3a shows ER positive surface epithelium while figure 3b shows PR positive surface epithelium on immunohistochemistry (strong nuclear positivity is seen for both ER/PR hormone receptors).

of Mullerian origin on the basis of the patient's clinical history, histopathological and immunohistochemical findings of the cyst. The patient reported uneventful recovery after four weeks of follow-up.

Discussion

Mesenteric cysts are more common in adult patients (mostly around 20-40 years of age); however, few subtypes like cystic lymphangiomas are more frequent in children as well as in the foetus.[4] No specific gender predilection is seen for mesenteric cysts formation. Most common sites of occurrence are the mesentery (60%), mesocolon (24%), and the retroperitoneum (14.5%), whereas in 1.5% of the cases the exact location remains unestablished.[5] The patients usually present with variable, non-specific symptoms depending upon the diameter of the cyst, its exact site, and the associated complications. However, the most commonly observed three categories of patients with mesenteric cysts are: (1) asymptomatic—diagnosed incidentally during physical examination, surgery, or routine imaging performed for other comorbidities and constitutes about (40–45%) of all cases; (2) nonspecific—abdominal pain and distension, along with nausea and vomiting, diarrhoea, constipation, and weight loss; and (3) acute abdominal symptoms—due to underlying inflammation, abscess and, rarely, rupture of the cyst wall. Different studies in the literature have reported six different types of mesenteric cysts: (1) cysts of lymphatic origin (simple lymphatic cyst and lymphangioma); (2) cysts of mesothelial origin (simple mesothelial cyst, benign cystic mesothelioma and malignant cystic mesothelioma); (3) cysts of enteric origin (enteric cyst and enteric duplication

cyst); (4) cysts of urogenital origin; (5) mature cystic teratoma (dermoid cysts); and (6) pseudocysts (infectious and traumatic cysts).[6]

The benign mesothelial cysts are lined by cuboidal epithelium whereas the underlying wall is made up of fibro-collagenous and fibro-vascular tissue.[3] The Mullerian cysts are urogenital in origin and in females the common sites of occurrence are vagina, uterus, and ovaries, while prostate and seminal vesicles are commonly involved in males. In addition, they may also be seen in lumbosacral and paravertebral region and least likely in the mediastinum. But mesentery is a very unusual site, and only a few cases have been reported up till now. Histologically, Mullerian cysts have cuboidal to low columnar epithelium. The underlying wall contains fibro-vascular tissue with few smooth muscle bundles.[2] Usually the lining of Mullerian cysts is positive for hormonal receptors like oestrogen and progesterone receptors and immunohistochemical stains including PAX-8 and WT1[7] which is consistent with the immunohistochemical findings of the present study.

Chylous cysts are lined by two to three layers of benign mesothelial cells. Few smooth muscle fibres are seen in underlying fibro-collagenous and fibro-vascular stroma.[8] The simple lymphatic cyst is characterised by thin lymphatic channels which are lined by flattened endothelium. The underlying wall and septa are made up of fibro-vascular tissue, along with lymphoid aggregates and few smooth muscle fibers.[4] The enteric cysts and enteric duplication cysts are lined by intestinal epithelium with smooth muscle bundles in the underlying wall.[9]

Pseudocysts do not have an epithelial lining and the wall is made of fibro-collagenous tissue containing cholesterol clefts, foamy cells, and may histologically resemble pancreatic pseudocysts. However, they are known as non-pancreatic pseudocyst to avoid any confusion with pancreatic pseudocysts.[10] Malignant cysts are very uncommon with a prevalence of less than 3%, and they usually represent other tumours initially suspected as mesenteric cysts.[11]

Ultrasound of the abdomen is usually the first radiological test performed which primarily identifies the presence of a cystic lesion; however, it cannot clearly explain the origin of the cyst and, hence, does not contribute in specifying diagnosis. Contrast-enhanced computerised tomography (CECT) of the abdomen specifically explains the site, extent, and consistency of the cyst, while a CT scan with contrast also rules out adhesion of adjacent intestine and other structures.[1,4] Although these cysts are benign, complications such as haemorrhage, infection, bowel obstruction, and recurrence can arise, particularly when they are not fully excised. In addition, excision may not be possible in some cases due to their close proximity with vital structures (e.g., portal vein, superior mesenteric vessels), leading to the death of young patients.[11]

Surgical resection of a mesenteric cyst is the treatment of choice which may be done either by laparoscopy or by open surgery with or without resection of the attached intestinal segment, which further depends on the presence of adhesions and compromised blood supply to the involved segment.[12] Until now, there are no established guidelines regarding the post-operative follow-up of these patients. However, it depends on various factors like morphological subtype, complete resectability, and a sudden increase in the size of the cyst as these features are associated with the risk of malignant neoplasm or an increased likelihood of further complications.[2]

Conclusion

Owing to its rarity, mesenteric cyst is challenging for the radiologist, pathologist, and surgeon alike, due to the difficulties encountered in its diagnosis and management. However, it must be included in the differential diagnosis and work up plans for intra-abdominal cystic lesions, in order to avoid unnecessary surgeries.

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SG: Concept, drafting and revision.
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MJ: Concept, drafting, data analysis and interpretation.
NN: Concept, design, revision and final approval.