Case Report

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Cystic lymphangioma of the jejunal mesentery in a young adult: a rare case report

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ABSTRACT

Lymphangioma is an uncommon congenital malformation of the lymphatic system that manifest as a benign tumor. They are most commonly seen in children and rarely in adults. Lymphangiomas in the peritoneal cavity are extremely rare in adults, comprising of less than 1% of all lymphangiomas. We report a case of 18-year-old healthy female who was diagnosed with cystic mesenteric lymphangioma of the small intestine on CT scan as a part of her routine health check-up. CT scan reported a large well-defined cystic mass in mesentery of the small intestine filled with a hyper dense fluid. The lesion had thin capsule and internal septa. Radiological diagnosis was confirmed intra-operatively on laparoscopy. The patient underwent laparotomy with resection of the cyst and the involved jejunum segment followed by end to end anastomosis. The specimen was sent for histo-pathological examination and the findings were consistent with the diagnosis of cystic lymphangioma of the jejunal mesentery. Cystic mesenteric lymphangiomas are rare entities in a clinical setting that can be easily missed on clinical examination or may present as asymptomatic disease. CT scan is the gold standard investigation for diagnosis of mesenteric lymphangiomas. The preferred surgical approach depends on the location of the lesion, the clinical features and the spread of the lesion to surrounding structures.

Keywords: Cystic lymphangioma, Jejunum, Small bowel mesentery

INTRODUCTION

Lymphangioma is an uncommon congenital malformation of the lymphatic system that manifest as a benign tumor. It is most commonly seen in children and rarely in adults. The common sites of occurrence of the tumor are in the head, neck and axilla. They are extremely rare in the peritoneal cavity. Lymphangioma of the peritoneal cavity in adults comprise of less than 1% of all lymphangiomas. ^{2,3}

We report a rare case of cystic lymphangioma of the jejunal mesentery in a young adult diagnosed incidentally on routine health check-up.

CASE REPORT

A case of 18-year-old healthy female who was diagnosed with mesenteric lymphangioma on ultrasonography of abdomen and pelvis as a part of her routine health check-up. She was asymptomatic and her past medical and family history were unremarkable. There was no history of past surgeries. Her general and systemic examinations did not detect any abnormality. Her laboratory studies were reported to be normal.

Modern CT scanners (MDCT) scan of whole abdomen revealed a large well-defined cystic mass in the mesentery. The size of lesion was 10.5 cm×10.0 cm×6.0 cm and was located in the left iliac fossa. The lesion was

seen in the mesenteric border of segment of proximal small bowel loops. The lesion had irregular outline with thin enhancing capsule and internal septa. On delayed scan there was minimal enhancement of internal septa. No solid component was noted. The cyst was filled with hyperdense fluid with a density of 20-25 HU. There was no evidence of bowel stricture or any obstructive changes. Mesenteric vessels of the involved loop appeared to traverse through the cystic lesion. Rest of the small bowel loops, ileoceacal junction and large bowel appeared normal. Mesenteric vessels revealed normal contrast enhancement. CT scan findings suggested cystic lymphangioma of the small bowel mesentery encasing a short segment of small bowel in the left iliac fossa (Figure 1).

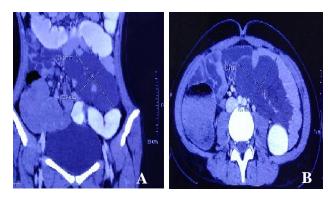


Figure 1: CT scan of the abdomen (A) coronal section (B) axial section showing an irregular cystic lesion in the left iliac fossa of size 10.2 cm×6.1 cm in the mesenteric border of segment of proximal small bowel loops.

Based on these findings the patient was planned to undergo diagnostic laparoscopy and to proceed further depending on the intra-operative findings. During laparoscopy the small intestine was examined using bowel grasper. A cystic lesion was visualized arising from the mesentery of jejunum 15 cm from the duodeno-jejunal flexure and extending for 25-30 cm from the duodeno-jejunal flexure. The lesion was not adhered to the intestinal wall or other organs. The ceacum appeared to be normal. Other abdominal and pelvic organs were normal.

Based on the above findings, laparotomy was done with excision of cystic lesion and resection of the involved bowel segment. End to end jejunal loop anastomosis was done in two layers. The second and third layer were anastomosed using vicryl 2-0 in continuous manner and the first and fourth layer were anastomosed using mersilk 2-0 in interrupted manner. A 28 no. ADK drain was placed at anastomotic site. The specimen was sent for histo-pathological examination.

Post operatively for the patient was kept nil-by-mouth and prescribed prophylactic broad-spectrum antibiotics, antacids, and analgesics. She was allowed to drink water and have liquid diet from the 5th post-operative day and the drain was removed on the seventh postoperative day. The post-operative period was unremarkable.

Histo-pathological findings of the specimen

Gross Examination: A specimen of small intestine 15 cm distal from duodenojejunal junction measuring 12.0 cm× 10.0 cm×4.0 cm was sent for examination (Figure 2). Two cystic swellings were identified arising from the mesentery. The first cyst measured 7.0 cm×3.0 cm and the second cyst measured 6.0 cm×5.0 cm. Cut section revealed chalky white fluid. The cysts were smooth and capsulated (Figure 3).



Figure 2: Gross specimen showing the resected segment of jejunum with two cystic lesions arising from the mesentery from either side of the jejunum.



Figure 3: Cut section of specimen showing cystic lesions arising from the mesentery of the resected jejunum. The cysts were smooth, capsulated and filled with chalky white fluid.

Microscopic examination: The specimen showed large irregular cystic spaces lined by flattened, bland endothelial cells. The surrounding scant stroma contained few lymphoid follicles, few congested blood vessels, smooth muscle aggregates, adipose tissue, aggregates of foamy macrophages and hemosiderin macrophages. Few cystic spaces (sinusoids) contained eosinophilic acellular material (lymph) and they reached upto muscularis

mucosa of bowel. Both the resection margins showed unremarkable histology. The section from adjacent normal bowel showed moderate lymphoplasmacytic infiltrate in lamina propria (Figure 4).

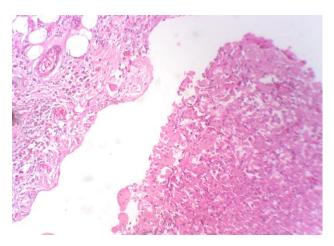


Figure 4: Microscopic image of the specimen (H and E stained).

These histo-pathological findings were consistent with the diagnosis of cystic lymphangioma of jejunal mesentery.

DISCUSSION

Mesenteric lymphangiomas are cystic lesions in nature and are therefore known as mesenteric cysts. These benign tumors have low incidence in the general population with intra-abdominal cystic lymphangiomas having an estimated incidence of<1/1,00,000 hospital admissions. ^{4,5}

In the abdomen, lymphangiomas are commonly located in small bowel mesentery (66% of cases) followed by ascending and transverse colon mesentery (33% cases). Less than 1% of cases have been reported in the mesentery of descending colon, sigmoid or rectum. They have a relatively asymptomatic onset, slow growth and a long disease course.

Lymphangiomas are commonly caused by congenital abnormality of the lymphatic system which leads to sequestration of lymphatic tissue during embryologic development.³ This theory explains why lymphangiomas are common in children and rare in adults. However, abdominal trauma, lymphatic obstruction, inflammatory process, surgery, or radiation therapy can also lead to the formation of such a tumor.^{8,9}

Lymphangiomas are classified as simple, cavernous, or cystic. Simple lymphangiomas are situated superficially in the skin and are composed of small thin-walled lymphatic vessels. Cavernous lymphangiomas are composed of dilated lymphatic vessels and lymphoid stroma, and have connection with spaces of various

normal adjacent lymphatics. Lastly, the cystic type consists of lymphatic spaces of various sizes that contains fascicles of smooth muscle and collagen bundles, but has no connection with adjacent normal lymphatics. However, cystic lymphangioma is not always clearly differentiated from cavernous type because the cystic type may also contain cavernous areas. 10,11

Clinically, mesenteric lymphangiomas usually present with non-specific symptoms such as abdominal pain, vomiting, and constipation. In cases such as ours, the patient may be completely asymptomatic. Hence, radiological investigations are gold standard for diagnosis of mesenteric lymphangiomas. The majority of lymphangiomas are discovered incidentally on imaging studies as a part of routine health check-up or for the investigation of unrelated clinical indications.¹²

On ultrasonography mesenteric lymphangiomas appear as cystic lesions, hypo echogenic, multilocular, with multiple internal septa and thick wall without vascularization on doppler study. While on CT scan which is the gold standard for diagnosis these appear as homogenous, hypo dense lesions with thin partitions that are enhanced when they are thick. 13,14

Surgical excision of the tumor is the gold standard method for treatment of lymphangiomas. The surgical technique used maybe laparotomy or laparoscopy depending on the location of the tumor, its size, involvement of the surrounding structures, presence of adhesions and general condition of the patient.

CONCLUSION

We report a case of cystic mesenteric lymphangioma in a young adult female presenting as an incidental finding in routine health checkup. The radiological findings were highly suggestive of cystic mesenteric lymphangioma which was confirmed intra-operatively. The patient was successfully treated by excision of the cyst with resection of the affected small intestine segment followed by end to end anastomosis.

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