Mesenteric lymphangioma: A rare intraabdominal finding in a pregnant woman

Abstract
Mesenteric cavernous lymphangioma is a rare benign tumor, not often described in literature. The etiopathogenesis of this tumor is largely unknown. Clinically, the presentation is variable and may be asymptomatic or present with subacute or acute abdomen. We describe here a case of 23-year-old pregnant woman with an asymptomatic abdominal mass which was incidentally detected after ultrasound examination during antenatal visit. The pregnancy was subsequently terminated and the patient underwent surgical excision of the tumor along with intestinal resection. The mass was pathologically diagnosed to be mesenteric cavernous lymphangioma.

Key words: Lymphangioma, mesenteric mass, pregnant woman

INTRODUCTION
Lymphangioma is a benign neoplasm which usually occurs in children, common sites being head, neck and axilla. These are considered to be congenital lymphatic malformations rather than true lymphatic neoplasms. Lymphangioma occurring in small bowel mesentery in an adult is very rare accounting for approximately 1% of lymphangiomas. Preoperative diagnosis is often difficult due to the clinical course that is frequently silent. Radiological investigations are a useful diagnostic tool, but definitive diagnosis is confirmed only by histopathology after a complete surgical resection. Surgical excision of the tumor is essential to prevent life-threatening complications and at times an extensive surgery involving adjacent organs may be required for complete resection.

CASE REPORT
A 23-year-old female presented with amenorrhea for 2 months. Pregnancy was suggested by a positive Gravindex test which is an agglutination inhibition test performed on a urine sample to detect pregnancy. The patient was further advised routine laboratory investigations and an ultrasound to confirm the pregnancy. Ultrasound abdomen showed a live fetus of 6 weeks maturity. In addition, a large non-homogenous lesion measuring 10.6 × 4.8 cm with ill-defined margins was seen in the infraumbilical region just below the abdominal wall. Ultrasound-guided fine-needle aspiration cytology (FNAC) was performed from the mass which revealed clusters of inflammatory cells and cyst macrophages in a background of proteinaceous material. A diagnosis of a benign cystic lesion was made. The patient was advised termination of pregnancy and surgical excision of the mass.

Preoperative CT and MRI evaluation was done. Contrast-enhanced CT examination revealed a well-marginated, spherical hypodense mass in the umbilical region. It was intraperitoneal in location and was encasing the mesenteric vessels (arrow) [Figure 1a]. The mass showed extension and streaking toward mesenteric surface of adjoining jejunal loops. The inferior limit of the tumor was at the pelvic brim. The mass measured approximately 8 × 10 × 15 cm in size. MR examination also confirmed the well-defined lesion and encasement of mesenteric vessels. The pelvic organs were not involved [Figure 1b]. The patient underwent laparotomy with excision of the mesenteric mass and adjoining bowel loop which was sent for histopathological examination.

Grossly, a segment of small intestine with attached mesenteric mass was received [Figure 2a]. Intestine measured 40 cm in length. In the mesenteric fat there was a nodular mass measuring 10 × 6 × 5 cm. The outer surface was smooth and congested. Cut section was lobulated, grey white, soft with tiny
cystic spaces [Figure 2b]. No attachment with the intestinal wall was seen. Multiple small lymph nodes were also isolated from the vicinity of the mass.

Microscopic examination showed a tumor composed of variable sized, interconnecting and cavernous channels separated by fibrous stroma and fat. Eosinophilic, proteinaceous material was noted in the lumen of these cystic spaces [Figure 3a]. A large number of lymphoid follicles and aggregates were dispersed in the stroma [Figure 3b]. Tumor was confined to the mesentery and not involving the adjacent small intestine. Sections from small intestine were essentially unremarkable. Four lymph nodes isolated showed sinusoidal dilatation.

Immunohistochemical staining with CD31 and CD34 highlighted the flat endothelial lining of the lymphatic channels [Figure 4a and b].

Final diagnosis of mesenteric cavernous lymphangioma was rendered. The post operative period was uneventful with the patient being discharged in a stable condition after 7 days.

**DISCUSSION**

Lymphangioma is a rare benign mass-forming entity characterized by numerous thin-walled lymphatic spaces. Although it can be encountered at any age, it usually manifests in the first few years of life. These are believed to arise from congenital malformation of lymphatic vessels leading to their sequestration during the embryonic period. However, other possible causes described are trauma, inflammation, surgery or radiation. Majority of cases are seen in the head and neck region (75%) followed by axilla (25%). Other sites like abdominal or mediastinal cavity are rare, accounting for approximately 5% of lymphangiomas. Among these, lymphangiomas of the small-bowel mesentery have been described in less than 1% of cases. Thus, the present case of a mesenteric lymphangioma in an adult female is quite rare.

Lymphangiomas are classified into three histologic types: Capillary (simple), cavernous, and cystic. The capillary (simple) type usually originates in the skin and consists of uniform small thin-walled lymphatic spaces. The cavernous type is composed of various sizes of dilated lymphatic spaces associated with lymphoid stroma and shows a connection with the adjacent normal lymphatic spaces. The cystic type consists of dilated lymphatic spaces of various sizes associated with collagen and smooth-muscle bundles in the stroma but lacks connection to the adjacent normal lymphatic spaces. The present case was of cavernous type characterized by variable sized dilated lymphatic spaces having lymphoid infiltrate in the stroma.

Mesenteric lymphangiomas can remain asymptomatic or present with abdominal pain, vomiting, and constipation. This can result in a diagnostic dilemma as these symptoms are common to many diseases of gastrointestinal tract. However, imaging modalities like ultrasonography, CT and MRI are useful tools for a proper diagnosis as was seen in our patient. These tests are also used to estimate the size of the tumor and its infiltration into the surrounding

---

**Figure 1:** (a) Contrast enhanced CT abdomen coronal reveals a hypodense, large spherical mass in the umbilical region which is encasing the mesenteric vessels (arrow). There is streaking and extension of disease into the mesenteric surface of adjoining jejunum. (b) MRI of lower abdomen and pelvis T1W showing a cystic lesion in the infraumbilical region. Pelvic organs are normal.

**Figure 2:** (a) Gross image of segment of small intestine with attached mesenteric mass showing bosselation and congestion externally. (b) Cut section is grey white with foci of congestion.

**Figure 3:** (a) Photomicrograph showing variable sized cavernous channels distended with proteinaceous material. Lymphoid follicles are interspersed within the septa (H and E, x100). (b) Higher magnification showing the lymphatic channels lined by flattened endothelial cells (H and E, x200).

**Figure 4:** (a) Immunohistochemical staining showing the endothelial cells to be positive for CD31 (x200). (b) Immunohistochemical staining showing the endothelial cells to be positive for CD34 (x400).
Mesenteric lymphangioma is a benign lesion, it often has life-threatening complications such as secondary infection, rupture with hemorrhage and volvulus or intestinal obstruction.[3,4] Surgical excision is the treatment of choice and in some instances extended laparoscopy-assisted subtotal resection with sclerosing therapy. JSPS 2005;10:746-51.

The coexistence of mesenteric lymphangiomas with pregnancy is very rare and to the best of our knowledge only three cases have been reported in English literature, all of whom had a favorable outcome. The clinical and management details of the cases are summarized in Table 1.[3,4,14]

CONCLUSION

Mesenteric lymphangioma in adults is a rare disease which may remain asymptomatic or present with life-threatening complications. Preoperative diagnostic tools are ultrasound and abdominal CT or MRI. Mesenteric lymphangioma should be considered in the differential diagnoses of patients found to have intra-abdominal cystic masses on imaging. Complete surgical resection is the ideal modality of treatment and definitive diagnosis is confirmed by histopathology.

REFERENCES


Source of Support: Nil, Conflict of Interest: None declared.