

# Case Report of Four Different Primary Mesenteric Neoplasms and Review of Literature

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## Abstract

**Introduction:** It is rare for primary tumors to arise from the mesentery. Lymphangiomas appear as congenital malformations of the lymphatic system or benign neoplasms as a large, thin-walled, often multilocular cyst. Mesenteric infiltration is common and during surgical treatment, adjustment of structures such as the bowel and resection of the spleen may be required. Cystic mesotheliomas are rare, benign tumors that originate from the peritoneal mesothelium and are more common in women. Mesenteric cysts are mostly benign and rare intra-abdominal tumors, and can be seen as occupying a large cyst. Malignant fibrous histiocytoma is a rare pleomorphic sarcoma that is more commonly encountered in men. After the extremities, the second most common areas to be affected are the retroperitoneum and peritoneal cavity.

**Case Presentation:** We encountered four cases of different primary mesenteric neoplasms that were operated at the Gazi Yasargil teaching and research hospital, department of general surgery, Diyarbakir, Turkey, between 2013 and 2014. We reviewed these primary mesenteric neoplasms and compared them with previous literature.

**Conclusions:** Primary mesenteric tumors are rare and mostly benign tumors. Complete surgical excision is necessary for all tumors and follow-up is necessary after surgery for malignant fibrous histiocytoma due to recurrence.

**Keywords:** Mesentery, Neoplasms, Lymphangiomas, Cystic Mesotheliomas, Mesenteric Cyst and Histiocytoma, Malignant Fibrous

## 1. Introduction

Primary tumors arising from the mesentery are rare. Most primary lesions are mesenchymal in origin and are mostly benign (1). Mesenchymal tumors arising from the intraperitoneal portion of the subperitoneal space may originate from the lymphatic, vascular, neuromuscular, or fatty tissue.

Lymphangiomas appear as congenital malformations of the lymphatic system or benign neoplasms and appear as large, thin-walled, often multilocular cysts. Mesenteric infiltration is common, and the adjustment of structures such as the bowel and resection of the spleen may be required during surgical treatment. Tumors of omental origin are less frequent (2).

Cystic mesotheliomas are benign tumors and originate from the peritoneal mesothelium. These rare tumors are more common in women. Tumors have no malignant potential, but recurrence is reported in 25 - 50% of cases. Typically, the lesion originates from the mesothelium, presents as grape-like clusters and consists of fibrous tissue. On computerized tomography, cystic mesothelioma

appears as peritoneal-based multilocular cystic mass or multiple thin-walled different unilocular cysts. Its characteristic feature is the involvement of the pelvic region. The lack of significant mass effect, calcification, or significant soft tissue component differentiates it from malignant metastatic diseases such as pseudomyxoma peritonei (3).

Retroperitoneal, omental, and mesenteric cysts are rare intra-abdominal tumors that occur in 1 in every 105,000 hospitalized patients. Many authors believe that these cysts derived from the same embryonic structure and have similar pathogenesis such as ectopic lymphatic tissue. These cystic tumors are mostly benign lesions, but can be seen as occupying large cysts affecting other organs (3).

Malignant fibrous histiocytoma (MFH) is a rare pleomorphic sarcoma. MFH is more common in men and the most common sarcoma in adults aged 60 - 70 years. MFH is most frequently observed in the extremities (49% of the lower extremities, 19% of the upper extremities). The second most commonly affected areas are the retroperitoneum (16%) and peritoneal cavity (5 - 10%). Lesions in visceral organs and gastrointestinal organs are extremely

rare (4). Recurrence of MFH is common. Ultrasonography can differentiate cystic mesenteric masses from solid ones. However, computed tomography (CT) is the most commonly used approach to diagnose tumors in the mesentery. Percutaneous imaging-guided or surgical biopsy is often necessary in the management of these tumors.

Here, we report the operation of four different cases of primary mesenteric neoplasms and compared these four cases with those reported in previous literature.

## 2. Case Presentation

Primary mesenteric neoplasms are very rare. We treated four different types of mesenteric primary cystic benign and solid malignant tumors at Gazi Yasargil teaching and research hospital, department of general surgery, Diyarbakır, Turkey, between Jan 2013 and May 2014. Table 1 summarizes the clinical CT and pathological findings of all the patients.

The first case involved a 45-year-old female patient who had been admitted to our hospital with the complaint of abdominal pain and nausea. Contrast-enhanced CT showed hypodense cysts (approximately 1 cm in size) in the central portion of the right kidney and macrolobulated hypodense mass (approximately  $5.5 \times 2.5 \text{ cm}^2$  in size) in the subhepatic space accompanied by fat density. Abdominal ultrasound examination showed lobulated cystic lesions (about  $64 \times 45 \text{ mm}^2$  in size), identified in the region adjacent to the lower pole of the right kidney. A gallbladder stone (approximately 5 mm in diameter) was observed in the lumen. The common bile duct and intrahepatic bile ducts were of normal width.

The patient underwent laparoscopic operation. After placing trocars at the appropriate sites and insufflation of the abdomen, the gallbladder was removed. Cystic masses, measuring  $9.5 \times 4.5 \text{ cm}^2$  in the subhepatic space were completely removed (Figures 1 and 2). Pathology reported cysts consistent with lymphangioma. The patient was discharged on postoperative day 7 without any complication. At the one year postoperative follow-up visit, the patient was well, with no complaints or signs of cystic mass at ultrasonography.

Our second case involved a 61-year-old woman who was admitted to our hospital because of abdominal distention and pain. The patient had a history of abdominal hysterectomy and appendectomy. MRI of the abdomen showed a cystic mass ( $8 \times 5 \text{ cm}^2$ ) in the inferior part of the right liver lobe. Laboratory values were normal with a cardiac ejection fraction of 60%. The patient underwent laparoscopic surgery. After placing trocars in the appropriate sites and insufflation of the abdomen, a multicystic mass ( $8 \times 5 \text{ cm}^2$ )



Figure 1. Laparoscopic View of the Cystic Abdominal Lymphangioma



Figure 2. Cystic Abdominal Lymphangioma

in the lower part of the liver and a few small cysts around the cecum were noticed (Figure 3).



Figure 3. Laparoscopic Appearance of Benign Cystic Mesothelioma

All cystic masses were completely removed. Pathological result revealed multilocular peritoneal inclusion cysts consistent with benign cystic mesothelioma. The patient

**Table 1.** Clinical, Computed Tomographic, and Pathological Findings<sup>a</sup>

	Age, years/sex	Clinical presentation	Location	MRI	US	CT	Histological subtypes
1	45/F	Abdominal pain, nausea	Subhepatic space, borderline to right kidney	(-)	Lobulated cystic lesions (about $6.4 \times 4.5 \text{ mm}^2$ in size)	(+) Hypodense cysts $5.5 \times 2.5 \text{ cm}^2$	Lymphangioma
2	61/F	Abdominal distention and pain	Below the lower part of the liver and around the cecum	cystic mass ( $8 \times 5 \text{ cm}^2$ )	Multicystic mass ( $8 \times 5 \text{ cm}^2$ ) in the lower part of the liver	(-)	Benign cystic mesothelioma
3	20/F	Distended abdomen and tenderness in the left lower quadrant	Left lower part of the mesentery, Greater omentum	Mesenteric multicystic lesions ( $33 \times 14 \text{ cm}^2$ )	Multicystic lesion $33 \times 14 \text{ cm}^2$	(-)	Mesenteric (omental) cyst
4	52/M	Abdominal mass, pain, distention	#: The mesenteric plan in the left paramedian area and & in the mesentery left side between the upper jejunum and left colon in the upper right side of the abdomen	(-)	Heterogeneous mass in left paramedian area	#: Heterogeneous mass approximately $15 \times 19 \times 12 \text{ cm}^3$ ; & CT of the abdomen revealed three heterogeneous masses of 22 cm, 10 cm, and 15 cm	Malignant fibrous histiocytoma

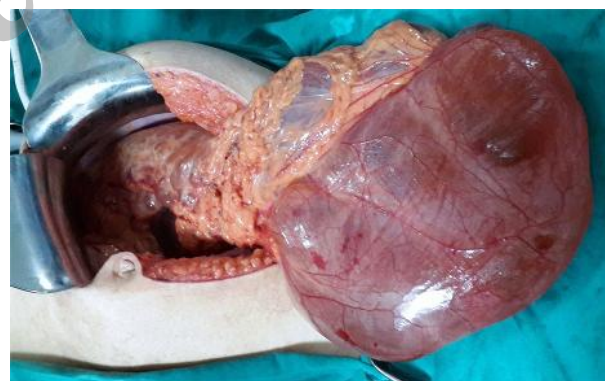
Abbreviations: CT, Computed Tomography; MRI, Magnetic Resonance Imaging; US, Ultrasonography.

<sup>a</sup>+, Present; -, Absent, #, during first operation; &, during second operation.

was discharged on postoperative day 5 without any complication. At the one year postoperative follow-up visit, the patient was well and ultrasonography showed no sign of cystic mesothelioma.

The third case involved a patient who was admitted to our hospital with the complaint of abdominal pain. Physical examination showed distended abdomen and tenderness in the left lower quadrant. Laboratory values were as follows: white blood cell count, 14510 cells/ $\mu\text{L}$ ; hemoglobin, 11.3 gm/dL; hematocrit, 33.1%; lactate dehydrogenase, 509 units/L; creatinine kinase, 970 units/L; and amylase, 123 units/L. A huge multicystic lesion with the dimensions  $33 \times 14 \text{ cm}^2$  appeared on ultrasonography. MRI showed mesenteric multicystic lesions ( $33 \times 14 \text{ cm}^2$ ) starting from the pelvis to the lower part of the left diaphragm around the spleen and stomach. After general anesthesia, the abdomen was opened through a midline abdominal incision. Upon exploration of the abdomen, a cystic mass starting from the pelvis, covering the entire abdomen, and reaching the left diaphragm, surrounding the stomach and spleen, was noticed (Figures 4 and 5). After clearly separating adhesion, the cystic lesions were completely removed and splenectomy was carried out. Pathology revealed a  $33 \times 14 \times 7.5 \text{ cm}^3$  cystic mass including large and small cysts consistent with mesenteric (omental) cysts. On postoperative day 6, the patient was discharged with no complications. At the 11-month postoperative follow-up visit, the patient was well and ultrasonog-

raphy showed no sign of mesenteric cysts, and the blood values were normal.

**Figure 4.** Intraoperative Picture of the Mesenteric (Omental) Cyst

Our fourth case involved a 52-year-old patient admitted to the clinic with complaint of an abdominal mass, pain, and distention. Contrast abdominal CT showed a heterogeneous mass approximately  $15 \times 19 \times 12 \text{ cm}^3$  in size in the mesenteric plan in the left paramedian area extending from the inferior border of the left kidney to the level of the iliac bifurcation, compressing intestinal structures.

<sup>18</sup>F-fluorodeoxyglucose (FDG) whole-body positron emission tomography (PET) was performed for diagnostic purposes. The solid cystic component was a  $141 \times 103$





**Figure 5.** Mesenteric (Omental) Cyst Including Spleen

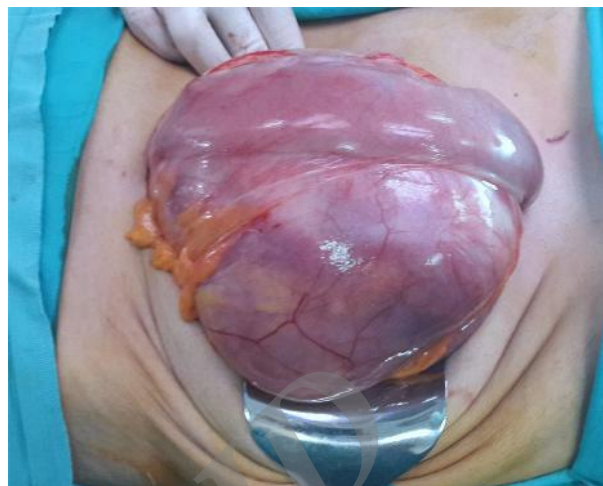
mm<sup>2</sup> mass with measured malignancy level increased FDG uptake with an SUVmax of 3.9 in the left middle abdominal quadrant between the peritoneal leaves. The FDG holding mass was compatible with the malignant mesenchymal tumor. The abdomen was explored through an old median skin incision. A mass measuring 20 × 18 cm<sup>2</sup> in the left lower quadrant of the abdomen was seen (Figure 6).



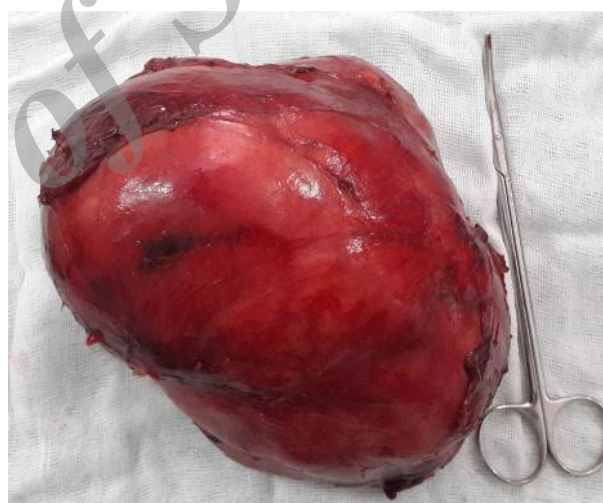
**Figure 6.** Operative Picture of a Malignant Fibrous Histiocytoma

The masses clung to the small bowel mesentery (Figure 7). Adhesions were carefully separated. The mass was completely excised (Figure 8).

Pathology reported a grayish purple-brown mass with the dimensions 22 × 18 × 18 cm<sup>3</sup> with a grayish purple and grayish yellow appearance in the cross-sectional face. The pathology was consistent with MFH. At postoperative day 7, the patient was discharged with no complications. The patient visited our clinic for the 12-month postoperative follow-up visit. Unfortunately, the disease recurred. CT of



**Figure 7.** The Malignant Fibrous Histiocytoma Clung to the Small Bowel Mesentery



**Figure 8.** Completely Excised Malignant Fibrous Histiocytoma

the abdomen revealed three masses in the mesentery measuring 22 cm and 10 cm in the left side between the upper jejunum and the left colon and 15 cm in the upper right side of abdomen. This patient was operated 3 months ago. After laparotomy, a mass adjacent to the upper jejunum and parallel to the left descending colon measuring 22 cm in diameter, a mass in the upper portion of this mass near to the treitz ligament measuring 8 cm in diameter, and a mass in the right side between the right colon and mesenteric superior artery and vein fixed to the mesentery were noticed. All the masses were removed with careful dissection while preserving the bowels and vascular structures. The patient was discharged on postoperative day 7 with no

complication. Pathology of the masses revealed malign fibrous histiocytoma. At the 4-month postoperative follow-up visit, the patient was well with no complaints. The patient received neither adjuvant chemotherapy nor radiation.

### 3. Discussion

Definition of peritoneal benign cystic mesothelioma (BCM): BCM has been previously described by Mennemeyer and Smith (5). BCM is a rare intra-abdominal tumor arising from pelvis that appears as a large multicystic mass. Thus far, approximately 140 cases have been described in the literature (6). Peritoneal mesotheliomas are divided into three subgroups: malignant peritoneal mesotheliomas, cystic mesotheliomas, and well-differentiated papillary mesotheliomas. There is an association between asbestos exposure and malignant peritoneal mesothelioma. Pathology of BMC: Cystic mesotheliomas consist of multiple grape-like clusters of mesothelium-lined cysts and may be seen in a single location. Thin-walled cysts of variable sizes are located in the greater omentum, pelvis, and under the right diaphragm (7). Some authors consider cystic mesothelioma as benign (8). In the literature, the disease appears as multilocular peritoneal inclusion cysts, multicystic peritoneal mesothelioma, and postoperative peritoneal cysts (6). Diagnostic approach of BMC: Abdominal ultrasound, CT, or MRI are employed to diagnose BMC. However, laparoscopy is also a good diagnostic tool. Laparoscopy provides biopsy and definitive treatment. In our case, all the cysts were removed laparoscopically. Treatment of BMC: Surgery with complete enucleation of all the cysts is the best treatment method. Follow-up of BMC: Peritoneal BCM has a high local recurrence rate (8) with a 40 - 55% recurrence rate in female patients and 33% relapse rate in male patients (9). In our patients, no recurrence was noticed up to the last follow-up. Recurrence is very high and malignant transformation is very small; therefore, heated intraperitoneal chemotherapy has been recommended after aggressive surgery (10).

Definition of abdominal cystic lymphangioma: Lymphangiomatosis is a very rare condition and is a subtype of lymphatic disease. A PubMed search for articles on lymphangiomatosis found 359 articles, of which 56 articles were about abdominal lymphangiomatosis in adult patients (11). Gastrointestinal and mesenteric lymphangioma are extremely rare in adults. The etiology of lymphangiomas is still unclear. Congenital dysplasia of the lymphatic tissue and abnormal development of lymphatic vessels during fetal life is considered in the etiology. Lymphangioma is usually classified as simple capillary, cavernous, and cystic according to the size of the lymph and

lymphatic walls on the nature of the field. Cavernous lymphangiomas are composed of dilated lymphatic vessels and lymphoid stroma and are associated with the normal adjacent lymphatic. Cavernous areas may have cystic lymphangiomas. The clinical symptoms of gastrointestinal and mesenteric lymphangiomas may vary according to tumor size and location and change from asymptomatic to acute abdominal symptoms such as obstruction or bleeding. Pathology of abdominal cystic lymphangioma: Majority of the abdominal cystic lymphangiomas were classified as benign lesions. Cystic mesotheliomas and lymphangiomas are benign and although they have few similarities, they show similar microscopic and gross features (12). Macroscopically, lymphangiomas appear as a cystic mass with partial septation and show histological characteristics of endothelial cells, dilation, variable amount of connective tissue, and communicating lymphatic channels containing a variable amount of connective tissue and smooth muscle fiber.

Diagnosis of abdominal cystic lymphangioma: A lymphangioma usually appears as a cystic mass on imaging studies such as ultrasound, CT, and MRI (13). Lymphangiomas appear as multilocular cyst walls by CT. Lymphangioma and cystic mesotheliomas have similar radiological findings. Some cystic lymphangiomas may appear as a solid mass on ultrasound and CT due to intracystic debris or hemorrhage or studies of intracystic or debris or microcystic structure of cavernous hemangiomas (13). However, it is not possible to understand significant differences between cystic and cavernous lymphangiomas. Intra-abdominal cystic lymphangiomas often appear as a cystic mass and are usually thin-walled, multiseptate, and with or without intracystic debris. Differential diagnosis of abdominal cystic lymphangioma: There are no specific findings distinguishing the origin or nature of intra-abdominal cysts. Presence of septa, wall thickness, and presence of calcification in the wall or bowel displacement help in the differential diagnosis. Abdominal cystic lymphangioma is common in adult women and requires multiple surgical procedures and tend to recur (3). Treatment of abdominal cystic lymphangioma: Complete surgical resection of the mass is the treatment of choice. Some require multiple surgical procedures and may recur (3). Follow-up of abdominal cystic lymphangioma: Follow-up is necessary. The prognosis is generally good, although there is a likelihood of tumor recurrence (13).

Definition of mesenteric and omental cysts: Although they are rare, mesenteric and omental cysts have been an attractive topic for researchers because of their uncertain pathogenesis and confusing terminology. Mesenteric cysts are estimated to be 3 - 10 times more common than omental cysts. Mesenteric and omental cysts have been accepted

as congenital abdominal lesions. However, only one-third of all published cases have occurred in children under the age of 15 years, and the majority of cases have been seen in adult patients. This may be because affected patients do not seek medical help until gross swelling occurs, because these tumors do not affect the patient's day-to-day activities (14). Treatment of mesenteric and omental cysts: The aim of surgical treatment is complete excision of the mass. During resection, adjacent structures sometimes need to be removed.

**Definition of mesenteric MFH:** Mesenteric MFHs are extremely rare and highly malignant neoplasms with early metastatic spread. Only 14 cases of mesenteric MFHs located in the upper mesentery have been reported in the English literature. Mesenteric MFHs typically present on CT as a poorly margined or well-circumscribed soft tissue mass, with hypodense areas due to necrosis. Pathology of MFH: Myxoid histology and tumor size are predictors for metastasis and recurrence. Myxoid tumors have a higher metastatic tendency than non-metastatic tumors (10-year metastasis rate: 13% versus 40%). Differential diagnosis of MFHs: These tumors must be differentiated from gastrointestinal stromal tumors, which account for most primary mesenchymal tumors of the gastrointestinal tract and leiomyomas, leiomyosarcomas, fibrosarcomas, and liposarcomas (4). Treatment of MFHs: The type of surgical treatment is related to the disease stage and tumor depth. Primary MFH is treated with surgical wide excision of the tumor with an aim for a tumor-free margin. Radiotherapy, chemotherapy, and immunotherapy are other treatments (4). However, the efficacy of adjuvant chemotherapy or radiation has not been clear in the case of retroperitoneal and visceral MFHs (15). Follow-up of malignant fibrous histiocytomas: MFHs are aggressive tumors with high rates of local recurrence and metastasis. The rates of local recurrence and distant metastasis are 44% and 42%, respectively (15). Distant metastases develop in the hematologic (30%) and lymphatic system (12%). Local recurrence and distant metastasis of the primary tumor increases with depth and size of the tumors. The most common site of metastases is the liver with a rate of 64 - 70%. Recurrence is not uncommon even if resection margins are tumor-free. Recurrence was seen in our case after one year of complete excision. Abdominal MFH shows worse prognosis than tumors of the extremities. The 5-year survival rate of patients with abdominal MFH is reportedly 14% (15). Therefore, follow-up is necessary due to tumor recurrence and metastasis, and CT is suitable for this purpose.

### 3.1. Conclusion

Primary mesenteric tumors are very rare. Lymphangiomas and cystic mesotheliomas are benign tumors.

Omental and mesenteric cysts are rare and mostly benign lesions. MFH is a rare sarcoma that is highly malignant. Ultrasonography and CT follow-up is necessary in all primary mesenteric neoplasms due to recurrence. Recurrence is high especially for MFHs.

### Footnotes

**Authors' Contribution:** Yusuf Yagmur was responsible for study design, data collection, management, and data analysis, and preparation, review, and approval of the manuscript. Sami Akbulut was responsible for data analysis and preparation, review, and approval of the manuscript. Serdar Gumus was responsible for data analysis and preparation, review, and approval of the manuscript.

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