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Primary leiomyosarcoma of the omentum presenting as an ovarian carcinoma, case report and review of the literature



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ABSTRACT

Primary omental leiomyosarcoma is a rare tumor. We report a case of successfully resected omental leiomyosarcoma whose presentation mimicked ovarian carcinoma. Symptoms of abdominal distension and discomfort that lasted 8 months followed by pain lead to a diagnosis of a large mass in the abdomen. Physical examination revealed a large, over 20 cm tumor, suspected to be of ovarian origin. A small amount of ascites was found on Computerized Tomography (CT) and ultrasound (US) scans. Total abdominal hysterectomy with bilateral salpingo-oophorectomy, omentectomy and tumor debulking procedure was planned. Laparotomy revealed normal uterus ovaries and tubes with a leiomyosarcoma of the omentum which was completely resected successfully. Only 26 cases of primary leiomyosarcoma of the omentum were previously described in the literature. A review of the literature is also presented.

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1. Introduction

Primary leiomyosarcoma of the greater omentum is a rare pathologic entity, and the literature includes only 27 cases including this one (Table 1). The pre-operative diagnosis of these tumors is difficult and the diagnosis is usually made post operatively.

We report a case of a 55 year old woman with clinical symptoms of abdominal distension and discomfort followed by pelvic pain. The omental origin of the tumor could not be identified using CT and US scans. We also review the literature.

2. Case report

A 55-year-old, previously fit and well woman, presented with increasing abdominal distension and discomfort for 8 months and pelvic pain that began a few days prior to presentation. A CT scan was performed as part of the evaluation and demonstrated a huge abdominal mass, measuring more than 20 cm in cross section in the left side of the abdomen. A small amount of ascites was also noted. The mass had some large draining vessels on the left side and there was also infiltration of the fat in the upper abdomen on the left side suggesting peritoneal disease. The mass was presumed to be ovarian in origin. Tumor markers were taken as part of the evaluation and her CA-125 levels

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were elevated at 527 U/ml, the levels of CEA, CA15-3 and CA19-9 were normal. Pelvic ultrasound scan was also performed and a 20×12 cm heterogeneous lower abdominal mass with cystic and solid components was found (Fig. 1). The uterus and contralateral ovary could not be well visualized. The nature of the lesion was uncertain according to the ultrasound scan. Ovarian carcinoma was suspected with high probability and the patient was consented for laparotomy, total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy and tumor debulking.

At the time of surgery, a $23 \times 20 \times 13$ cm irregular mass arising from the omentum and appearing to be parasitic in nature was found, the mass had extensive recruitment of huge vessels from the omentum. The pelvis was obliterated by adhesions consistent with old endometriosis. The ovaries were small and adherent to the posterior uterus. The findings were not consistent with gynecologic malignancy. The liver, diaphragmatic surface, and all peritoneal surfaces were normal. The small bowel and colon were normal and a frozen section analysis of the mass suggested sarcoma. Omentectomy was performed and the tumor removed intact, no further omental spread was noted. Following division of adhesions, total abdominal hysterectomy and bilateral salpingo-oophorectomy were also performed.

Histologically the tumor had the classical appearance of a leiomyosarcoma (Fig. 2). The tumor seemed to be arising from the smooth muscle in blood vessel walls in the omentum. Histopathologic examination revealed a multinodular but smooth outer surface of the tumor and foci of fleshy and pale cream-yellow with mucoid/mixoid areas underneath. The microscopic examination confirmed the

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Table 1

Review of previously reported cases of primary omental leiomyosarcoma, diagnosis, management and outcome.

Number (reference)	Age (years)	Sex	Symptoms	Imaging	Preoperative diagnosis	Tumor size and Spread	Treatment	Outcome
1 (Stout et al., 1963)	38	М	Abdominal mass and pain	NA	Abdominal mass	Implants on small bowel and peritoneum	Biopsy	Died 48 h post-op.
2 (Stout et al., 1963)	29	F	Mass, uterine bleeding, abdominal	NA	Uterine bleeding, abdominal mass	Tumor described as 'Huge' 3 omental tumors 3 cm, 5 cm and 6 cm. Peritoneal implants.	Hysterectomy. Excision of tumors	Died 18 months post-op.
3 (Stout et al., 1963)	26	F	Pain	NA	Uterine bleeding, abdominal distension	20 cm omental tumor, fibroid uterus, hemoserous ascites	Hysterectomy. Excision of tumors	Died 36 h post-op. due to PE
4 (Weinberger & Ahmed, 1997)	68	F	Abdominal mass	NA	Abdominal mass	NA	Omentectomy	Alive 2.5 years follow-up
5 (Weinberger & Ahmed,	80	М	Pain	NA	Abdominal mass	NA	Omentectomy	Died 6 months post-op.
1997) 6 (Tanimura et al., 1980)	52	F	Abdominal mass	NA	Abdominal mass	$11 \times 26 \times 15$ cm	Excision	Alive 3 years follow-up
7 (Tanimura et al., 1980)	46	М	Pain	NA	Epigastric pain	$8 \times 10 \times 10$ cm, spread to stomach	Excision	Alive 7 years follow-up
8 (Fattar et al., 1981)	52	Μ	Abdominal mass	Angiography Right gastroepiploic artery	Abdo	4.2 kg tumor with peritoneal seeding	Excision	NA
9 (Dixon et al., 1984)	85	М	Fullness	Angiography – normal	Hemorrhagic ascites	6 cm	No treatment	Died within 2 days from presentation
10 (Scwartz et al., 1991)	40	М	Pain	CT – mass	Abdominal mass	10 cm	Excision, omentectomy	Alive 1.5 years follow-up
11 (Lee et al., 1991)	42	F	Abdominal mass	US CT	Abdominal mass	20 cm	NA	NA
12 (Lee et al., 1991)	60	М	Abdominal mass	US CT	Abdominal mass	20 cm	NA	NA
13 (Lee et al., 1991)	55	М	Abdominal mass	US CT	Abdominal mass	10 cm	NA	NA
14 (Langlieb et al., 1992)	46	F	Abdominal mass + pain	СТ	Ovarian carcinoma	20 cm	Excision, hysterectomy + BSO, omentectomy	NA
15 (Mahon et al., 1993)	51	М	Abdominal mass	CT				
16 (Ishida et al., 1999 Mar)	44	Μ	Abdominal mass	CT US Angiography - gastroepiploic artery	Omental tumor	28 × 25 cm	Excision, omentectomy	Alive 6 months follow-up
17 (Tanimura et al., 1980)	48	М	Abdominal mass			50 g, greater omentum	Excision	Died (post-op)
18 (Tanimura et al., 1980)	29	F	NA			$6\times5\times3$ cm, greater omentum	Excision	Recurrence, died
19 (Tanimura et al., 1980)		F	Abdominal distress			20 cm, greater omentum	Excision	Died (post-op)
20 (Tanimura et al., 1980)	32	М	Abdominal distress			6×4 cm, gastrohepatic omentum	Excision	Died-metastasis
21 (Tanimura et al., 1980)	70	F	Abdominal mass			$22\times14\times13$ cm, gastrohepatic omentum	Excision	Died-metastasis
22 (Tanimura et al., 1980)	55	М	Abdominal distension			Multiple, greater omentum	Excision	Died-metastasis
23 (Tanimura et al., 1980)	43	М	Abdominal mass			$22\times19\times12$ cm, gastrohepatic omentum	Excision	NA
24 (Tsurumi et al., 1991)	59	М	Abdominal mass	US, CT, angiography, laparoscopy		Greater omentum	Excision	Alive
25 (Kimura et al., 1997)	58	М	Pain, nausea	I		Lesser sack	Excision	
26 (Koga et al., 2002)	63	F	Abdominal mass		Leiomyosarcoma	$12.5 \times 9 \times 8$ greater omentum and 6 liver metastases	Excision + chemotherapy	Alive
27 (our case)	55	F	Abdominal mass and pain	US, CT	Ovarian carcinoma	$23 \times 20 \times 13$ cm	Excision	Alive

diagnosis of sarcoma with moderately cellular interlacing fascicles of spindle cells with a high degree of mitotic figures and atypical forms along with areas of mixoid change and coagulative tumor necrosis. Immunohistochemical staining was diffusely positive for desmin and smooth muscle actin, and strongly negative for S100 and CD34 in keeping with leiomyosarcoma. The rest of the omentum was free of tumor. The uterus, cervix, ovaries and fallopian tubes showed no evidence of disease, a positron emission tomography (PET) scan demonstrated no further suspicious lesions. The case was reviewed at the gynecologic on-cology tumor board and also at the specialized sarcoma unit tumor board meetings and both advised no adjuvant chemotherapy or radiotherapy.

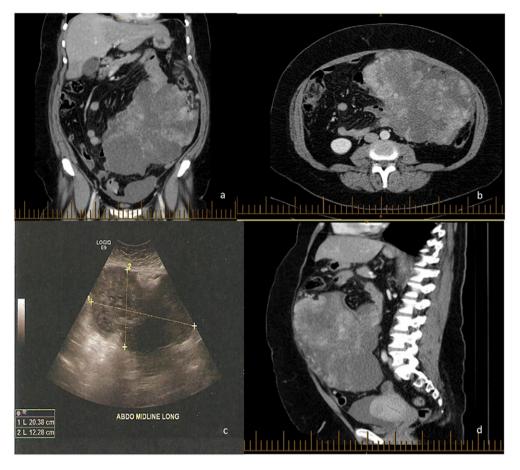


Fig. 1. Computerized Tomography (CT) imaging of leiomyosarcoma (a, b, d) and ultra sound (US) scan (c) of primary leiomyosarcoma of the omentum.

3. Discussion

Leiomyosarcoma of the omentum is a rare condition and can present in various ways. In this case a large pelvic tumor with omental involvement, ascites and elevated CA125 was discovered in an otherwise healthy patient.

Ultrasound scan was able to detect the tumor and showed accurately the internal structure of the lesion, nevertheless, it failed to determine the site of origin. This limitation of US may be attributed to the presence of bowel gas and attenuation of the US beam passing through a huge tumor, both factors prevent precise observation of the anatomic relationship between the lesion and the neighboring organs (Ishida et al., 1999). A CT scan can sometimes determine the omental origin of the tumor. Ishida and Ishida (1998) stated it to be the imaging of choice for greater omental tumors. A gastro-colic separation resulting from a tumor in the omentum can sometimes be seen (Fattar et al., 1981) and their CT appearance is usually multilobulated, flat, and pancakelike, with enhancing solid and multicystic densities. However, in this case the presentation, US and CT diagnosis were suspected to represent ovarian carcinoma. There is one other case described in the literature in which leiomyosarcoma was initially diagnosed as an ovarian malignancy (Langlieb et al., 1992). Ascites and elevated CA-125 that are sometimes associated with this tumor (Dixon et al., 1984; Langlieb et al., 1992) can suggest the initial diagnosis of ovarian carcinoma.

Angiography was described as useful in the diagnosis of greater omental tumors (Dixon et al., 1984; Fattar et al., 1981; Ishida et al., 1999). A diagnosis of a greater omental tumor can be suspected when a feeding artery to the tumor originates from the omental blood supply. The major arterial blood supply of the greater omentum is largely from the right and left gastroepiploic arteries, which derive from the gastroduodenal and splenic arteries (Sivak, 1992). A very vascular lesion with neovascularity, is more likely to represent a malignant tumor (Ishida et al., 1999).

Leiomyosarcoma is but one of many greater omental tumors. Reported primary tumors of the omentum, include leiomyosarcoma, fibrosarcoma, hemangiopericytoma, spindle cell sarcoma, liposarcoma, leiomyoma, lipoma, desmoid tumor, fibroma, mesothelioma, and others (Fattar et al., 1981; Ishida et al., 1999; Stout et al., 1963; Weinberger and Ahmed, 1997) They derive from different elements in the greater omentum which is composed mainly of fat but contains various tissues- such as vessels and lymphatics. Nevertheless, all of these primary omental tumors are very rare.

The frequency of omental leiomyosarcoma is uncertain. Altogether 27 cases (including this one) have been reported in the literature up to date. Thirteen cases were published between 1934 and 1979 and reviewed by Tanimura et al. (1980). From 1980 to the present, another 14 cases have been reported. Only one of the previously reported cases was suspected to be ovarian carcinoma at the outset (Langlieb et al., 1992).

The median age of patients with Leiomyosarcoma of the omentum in the cases published in the literature was 51 years (range: 26–85, SD: 15.3). The tumor is slightly more common among males (16 patients, 59.2%) and females (11 patients, 40.7%).

Ishida et al. (1999) has reviewed the cases from 1963 to 1999 and found a correlation between a symptom free mass and a better prognosis. This correlation is not as clear when adding the cases published before 1963, although much of the details regarding these cases are unavailable to us. Also, due to the limitations of imaging techniques in that era, early diagnosis was less likely.

Although these tumors are very rare, a diagnosis of leiomyosarcoma should be considered in a patient with an abdominal mass or distention and imaging studies demonstrating a huge mass with a central cystic

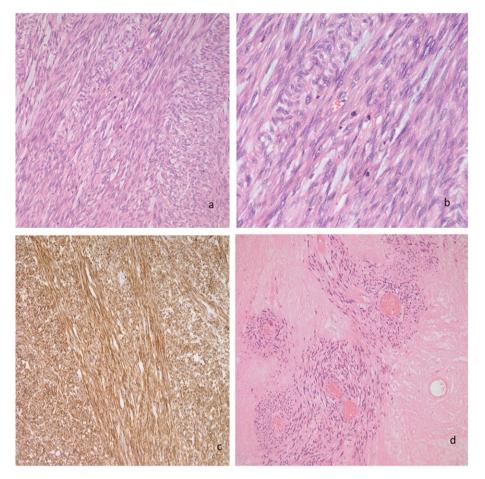


Fig. 2. Histology of primary leiomyosarcoma of the omentum. a: Tumor displaying long intersecting fascicles of spindle cells (H&E). b: Mitotic figures are readily identified within the tumor (H&E). c: Desmin immunohistochemical staining shows strong diffuse positivity, confirming smooth muscle differentiation. d: Abnormal blood vessels with thickened walls containing the same malignant spindle cells (H&E).

area in the greater omentum. The CT scan is probably more useful than US in the diagnosis of these cases and angiography may also prove useful demonstrating omental blood supply to the tumor. An accurate diagnosis can only be achieved with a histopathologic examination of the tumor (Ishida et al., 1999).

Complete surgical excision of thee tumors if feasible is probably the best option and can lead to long periods of disease free survival.

4. Conclusion

We report a case of a leiomyosarcoma of the greater omentum, presenting as ovarian carcinoma. Complete surgical excision of the tumor was performed and the patient recovered well. These tumors are very rare and the accurate determination of their precise anatomic location is usually difficult. The method of choice for imaging these tumors is CT scan with or without angiography. Complete surgical excision of these tumors can lead to long term survival.

Conflict of interest statement

The authors declare that there are no conflicts of interest.

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