Solitary Fibrous Tumor of the Omentum: Presentation of a Case and Literature Review

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ABSTRACT

Solitary fibrous tumor (SFT) and hemangiopericytoma (HPC) were considered, since their firsts description in the literature, as separate entities. The World Health Organization (WHO) classification of soft tissue tumors in 2013 declared the term HPC obsolete, and considered these lesions as features of the extrapleural SFT category. Herein we present a rare case of SFT originating from the great omentum. A 68 years old woman was admitted to our hospital with acute abdominal pain. Computed tomography revealed a 142 x 102 x 100 mm solid mass located in the pelvis, that simulated an adnexal lesion. An explorative laparotomy was performed, and a mass of the great omentum with a significant vascular pedicle arising from a branch of the left gastroepiploic artery was revealed. The tumor was completely resected. Microscopically it was composed by non-organized and spindle-shaped cells exhibiting atypical nuclei, arranged in short fascicles, and was diagnosed as. An extensive search was conducted in public scientific databases for published articles on the topic, with the aim to comprehensively describe the demographic, clinical, pathological and prognostic features of SFT; 60 previous cases have been identified and reviewed.

Key words: omentum, solitary fibrous tumor, hemangiopericytoma, mesenchymal tumor, SFT/HPC

INTRODUCTION

The greater omentum is an anterior vascular fatty apron-like fold supporting the abdominal viscera protecting them against tumors and infections. The most common malignancies of the greater omentum are due to carcinomatosis, secondary to peritoneal or hematogenous spread of digestive (colorectal, stomach, pancreas) or ovarian tumors. Solitary fibrous tumor (SFT) is a mesenchymal neoplasm that generally grows in the pleura, and occasionally in other body districts, including subcutaneous tissue, head and neck (especially the orbit), thoracic wall, mediastinum, pericardium, and retroperitoneum. Other reported locations include the meninges, spinal cord, periosteum, as well as organs like the salivary glands, lung, thyroid, liver, gastro-intestinal tract, adrenals, urinary bladder, prostate, spermatic cord and testes, and can be both benign and malignant (1). Only a few SFT cases involving the omentum have

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Abbreviations: SFT - solitary fibrous tumor; HPC - hemangiopericytoma; CECT - contrast-enhanced computed tomography; TKIs - tyrosin kinase inhibitors;

Received: 24.01.2020 Accepted: 10.03.2020

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been reported in the scientific literature to date. We herein report a rare case of omental SFT.

CASE REPORT

A 68 years old Caucasian woman, with no remarkable medical history, was referred to the University Hospital of Udine, Italy, for abdominal pain lasting 48 hours, with no further symptoms such as fever, stomach-ache, nausea or vomiting. Clinical examination showed a plethoric and soft abdomen, with no remarkable signs, but with a tender palpable mass moderately painful in the right lower abdominal quadrant. Laboratory data showed high white blood cell count (13200/µL) and C-reactive protein (41 mg/L). A contrast-enhanced computed tomography (CECT) scan of the abdomen and pelvis was performed, and showed a 142x102x100 mm mass occupying large part of the lower abdominal cavity, compressing the bladder and uterus, and resembling an adnexal lesion (figures 1 and 2). The mass showed low signal density, with regular and well defined borders, and after contrast injection, it was clearly enhanced, showing a mixed pattern of well vascularized and apparently necrotic areas. The mass drained into a dilated omental venous plexus. Neither vascular thrombosis, nor spread to adjacent organs were detected.

An explorative laparotomy was performed, and a large tumor originating from the greater omentum was found. The lesion, partly surrounded by omentum,

was tightly adherent with the inferior curve of the stomach. A dilated artery apparently originating from the left gastroepiploic artery, which seemed to feed the tumor, and an abundant and expanded blood vessel network, were present along the surface of the mass, leading to a 100 ml blood loss during excision. The lesion was successfully isolated and excised in toto with no technical difficulties and without resection of adjacent organs. The postoperative course of the patient was uneventful, and she was discharged five days after surgery. The patient gave her informed consent for all the medical procedures, and for the use of her anonymous clinical data for research purposes.

At gross pathological examination, the tumor measured 140x95x80mm in size, and was wellcircumscribed, solid, multilobulated, beige-pink in colour, with a 55mm haemorrhagic area (figure 3). Microscopically, it was composed of non-organized spindle-shaped cells, divided by branching vessels with intravascular thrombosis and multiple ischemic areas. No remarkable atypia was detected. Few mitotic figures (<3/10 high power field) were present. No immunohistochemical staining for EMA, CD99, cytokeratine, desmin, CD10, CD117 and DOG-1 was found, while staining positivity for CD34, bcl-2 and smooth muscle actin (SMA) was detected, leading to the diagnosis of a SFT. The patient was regularly followed-up with periodic imaging for the last four years, and no recurrences occurred.

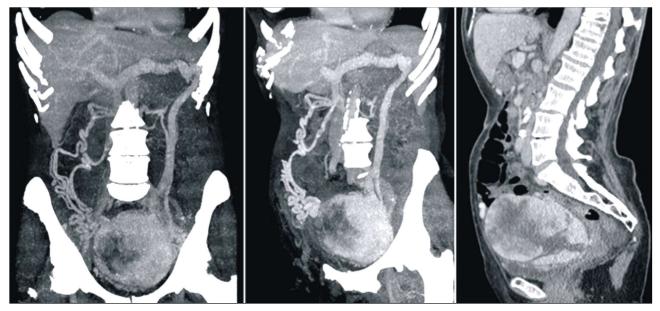


Figure 1 - Abdominal computed tomography scan showing the pelvic omental SFT



Figure 2 - Abdominal computed tomography scan showing a hypervascularized and heterogeneous solid tumor in the pelvis, with vascular feeding from the left gastroepiploic vessels

DISCUSSION

The first description of SFT dates back in 1931 (2) as a neoplasm originating from the pleura. The pleural sheets are most commonly affected by malignant mesothelioma, and represent the most common site of occurrence of SFT (3). It has been, indeed, hypothesized that SFT originates from sub-mesothelial mesenchymal cells (4). Extrapleural sites of origin, like the peritoneum, mediastinum, limbs, orbits and parotid glands have been subsequently described, and are less frequent but associated with particular clinico-pathological implications and higher risk of malignancy. Hemangiopericytoma (HPC), on the other hand, was first described by Stout and Murray in 1942 as a rare tumor of Zimmermann's pericytes (5). Although this tumor can arise anywhere, the muscles of the lower limbs, the pelvic fossa, and the retroperitoneum are the predominant sites of origin described. The fourth edition of the World Health Organization (WHO) Classification of Tumours of Soft Tissue and Bone published in February 2013 (6) erased these two entities, which are since then considered a unique tumor occurring commonly at all the sites of origin mentioned. Nevertheless, some pathologists continue

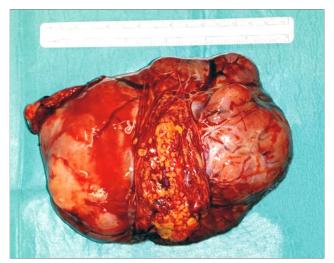


Figure 3 - Gross appearance of the resected SFT: a well-encapsulated pink and smooth solid mass, measuring approximately 14 cm

to maintain the old classification, despite the morphological and pathological features of the lesions are indeed related.

To date, only a few cases of SFT have been reported. We conducted an extensive electronic article search on PubMed and Scopus databases with the aim to identify published reports on SFT. The following keywords were used for research: "solitary fibrous tumor", "solitary fibrous tumour", "hemangiopericytoma", "haemangiopericytoma", "great omentum", "greater omentum", "omentum" and "omental" both as single terms or in combination. All the results were screened by two researchers (CB, AU), regardless of the reporting language, for cases with demographic, clinical and pathological information available. Subsequently, a cross-check of the reference lists of selected articles was performed to find any missed papers.

This way, there were 60 cases identified in 57 articles (7-63), a number consistently higher than those reported in previous reviews. The main demographic, clinical, pathological and immunohistochemical data of the cases enrolled are summarized in *table 1*. SFT did not show any gender predilection. Most cases occured during the 5th and 6th decades of life, and no environmental factors have been identified to increase the risk of SFT (64). The mean age in the whole cohort was 51 (range 24-92) years, and 29 patients were males (51%, *table 1*). In five patients the age or gender were not mentioned.

Patients with intra-abdominal SFT most commonly had a relatively indolent behaviour, and were often affected by a palpable abdominal mass or abdominal pain, vomiting and weight loss (65). Information about the personal clinical history was available in 52 cases; only in 9 (15%) cases the patients were totally asymptomatic, while abdominal or pelvic pain (41%), abdominal-pelvic mass (21%) and vomiting (7%) were the clinical manifestations most frequently encountered (*table 1*). The clinical symptoms were

Table 1 - Main demographic, clinical and immunohistochemical findings of SFT of the greater omentum as depicted in the current literature

Author (citation)	Year	Age/Gender	Symptoms/signs	IHC
Stout et al. (7) AUTOPSY	1943	92/M	abdominal mass	NA
Stout et al. (8)	1963	63/M	abdominal mass with pain	NA
Stout et al. (8)	1963	57/F	NA	NA
Stout et al. (8)	1963	64/M	abdominal pain, nausea	NA
Lortat-Jacob et al. (9)	1966	NA/M	abdominal mass, constipation	NA
Goldberg et al. (10)	1968	30/F	abdomnial pain	NA
Meyer et al. (11)	1976	55/M	abdominal mass, constipation	Reticulin+
Krejczy et al. (12)	1978	NA/NA	NA	NA
Alusik et al. (13)	1978	NA/NA	NA	NA
Harder et al. (14)	1983	37/M	abdominal pain	NA
Karabanov and Safiullin (15)	1989	61/F	abdominal mass	NA
Imachi et al. (16)	1990	62/F	abdominal distension with pain	NA
Schwartz et al. (17)	1991	40/M	abdominal mass with pain, wheight loss	NA
Cajano et al. (18)	1995	NA/NA	NA	NA
Bertolotto et al. (19)	1996	33/F	No	NA
Pozharliev et al. (20)	1997	45/M	abdomnial pain, atshenia	NA
Watanabe et al. (21)	1998	66/M	No	CD34+
Borgmann et al. (22)	1999	32/F	abdominal mass	NA
Borgmann et al. (22)	1999	57/F	abdominal mass	NA
Rao et al. (23)	2000	67/M	abdominal lump	NA
Kaneko et al. (24)	2003	70/F	lower abdominal mass grown over the past year	NA
Bovino et al. (25)	2003	46/F	severe upper abdominal pain, nausea, vomiting	vimentin+, keratin-, LCA-, S100-
Patriti et al. (26)	2004	24/M	lower abdominal pain, diarrhea, fever	CD34+, bcl-2+, pancytokeratin-
Ahmad et al. (27)	2004	74/F	abdominal distension, weight loss, diarrhea, vomiting	CD34+, vimentin+, SMA+, desmin+, Factor VIII+, S100-, cytokeratin-, CEA-
Crusco et al. (28)	2005	28/M	acute pain in the lower left quadrant	NA
Kim et al. (29)	2005	60/F	NA	NA
Piazza et al. (30)	2005	28/F	acute pelvic pain	NA
Sukharev et al. (31)	2006	NA/NA	NA	NA
Shiba et al. (32)	2007	41/F	epigastric pain	CD34+, factor XIIIa+, HLA-DR+
Slupski et al. (33)	2007	61/M	left lumbar pain	NA
Salem et al. (34)	2008	60/M	periumbilical pain, weight loss	CD34+, CD99+, SMA-, desmin-, S100-, C-kit -
Chatterjee et al. (35)	2008	41/M	NA	CD34+
Peixoto Callejo (36)	2009	44/M	abdominal tumour, anorexia, weight loss	CD34+, CD117-
Mosquera and Fletcher (37)	2009	40/M	lower abdominal pain	CD34+, p16+, p53+, EMA-, AE1/AE3-
Prakash et al. (38)	2009	45/F	lower abdominal pain	NA
Kucuk et al. (39)	2009	70/M	abdominal diffuse pain, nause, vomiting	CD34+, vimentin+

Author (citation)	Year	Age/Gender	Symptoms/signs	IHC
Maassarani et al. (40)	2010	63/F	NA	NA
Morris-Stiff et al. (41)	2011	68/M	No	CD34+
Garbin et al. (42)	2011	27/F	No	NA
Furukawa et al (43) (AUTOPSY)		69/M	NA	CD34+, vimentin+, SMA+, type IV collagen+, S-100-, cytokeratin-
Uemura et al. (44)	2012	48/M	painless mass in the left inguinoscrotal area	CD34+, CD99+, desmin-, S100-, SMA-, bcl-2-
Zong et al. (45)	2012	29/M	epigastric discomfort and compression, weight loss	CD34+, CD99-, bcl-2+, SMA+, vimentin+, CD117-, CD68-, cytokeratin-, calretinin-, desmin-, EMA-, F8-, S100-
Virgilio et al. (46)	2014	74/M	No	CD34+, bcl-2+
Becker et al. (47)	2014	41/F	distended abdomen, early satiety, postprandial vomiting	CD34 +, bcl-2+, vimentin+
Osawa et al. (48)	2014	32/F	irregular vaginal bleeding	CD34+, bcl-2 +, SMA+, S-100-, c-kit -
Harada et al. (49)	2014	62/F	vaginal discharge	CD34+, CD99+, bcl-2+, vimentin+, p16+, p53+, CD10+, PR+, S100+, c-kit+, EMA+, cytokeratin AE1/AE3+, SMA-, desmin-, D2-40 -, calretinin-, ER-, CD31-
Sato et al. (50)	2014	85/F	hypogastric mass	NA
Senda et al. (51)	2014	45/F	No	CD34+, bcl-2+, c-kit+, S-100-, desmin-
Cazejust et al. (52)	2015	68/F	left subcostal pain	CD34+, bcl-2+, C-kit -, DOG1-
Urabe et al. (53)	2015	52/M	No	CD34+, STAT6+, C-kit-, S100 -, desmin -, 1A4-
Moszynski et al. (54)	2016	29/F	pelvic pain, loss of appetite, bloating	CD34+
Jaber et al. (55)	2016	69/F	lower abdomen pain	vimentin+, CD34+, CD31+, reticulin+, keratin -, EMA-
Archid et al. (56)	2016	25/M	lower abdominal pain	CD 34+, CD99+, AE1/3-, desmin-, CD31-, CD117-, DOG1 -, S100- MIB1 5%
Rodriguez Tarrega et al. (57)	2016	34/F	No	CD34+, CD99+, beta catenin+, SMA-, desmin-, kit-, DOG1-
Michiura et al. (58)	2016	36/M	cough, abdominal mass	CD34+, bcl-2+, CD99+, p53+, S-100-, a-SMA-, c-kit-, desmin-
Bushira S.S. (59)	2017	45/M	abdominal pain and swelling	CD34+, factor-IIIa+, HLA-DR+
Yousefi et al. (60)	2018	24/F	lower abdominal pain	CD34+, CD99+, cytokeratin-, LCA-, CD117-, Synaptophysine-, Calretinine-, Inhibin-
Vasdeki et al. (61)	2018	72/M	recurrent mass of the anterior abdominal wall	CD34+, CD99+, vimentin+,
Jung and Bae (62)	2019	57/M	No	CD34+, STAT 6+
Suzuki et al. (63)	2019	45/F	abdominal pain	CD34+, bcl-2+, STAT6+, CD99+, c-kit-, S-100-, desmin-

Table 1 - Main demographic, clinical and immunohistochemical findings of SFT of the greater omentum as depicted in the current literature (continuation)

EMA: epithelial membrane antigen; ER: estrogen receptor; F: female; HIC: immunohistochemistry HPC: hemangiopericytoma; LCA: leukocyte common antigen; M: male; NA: not available; PR: progesterone receptor; SFT: solitary fibrous tumor; SMA: smooth muscle actin often attributable to a mass-compression effect on the surrounding anatomic structures. In addition, three cases manifested with haemoperitoneum (26,28,39), while five lesions mimicked an ovarian tumor (16,27,54,57,60).

The imaging techniques most frequently employed were roentgenograms and CECT. SFT tend to be welldefined, ovoid, and heterogeneously enhanced lesions in imaging. CECT and magnetic resonance imaging (MRI) appearance of abdominal and pelvic SFTs commonly consists in large, well-defined, ovoid, moderate enhancing masses, with heterogeneous CT attenuation or MRI signal intensity due to variable degrees of necrosis, haemorrhage and/or cystic evolution (65,66). Such non-specific imaging features lead to a wide spectrum of differential diagnoses to consider, including mainly other highly vascularized and/or fibrous-rich tumours such as leiomyosarcoma, neurogenic tumour, lymphoma, malignant fibrous histocytoma and mesothelioma. (65). The presence of a prominent vascular pedicle, although non-specific, has been described as a useful diagnostic feature of SFTs, and has been reported in 35% to 100% of the cases (67).

Histologic features that define SFT as a malignant neoplasm have been proposed, but no unanimous criteria have been established. Zong et al. (45) suggested a risk assessment algorithm to predict SFT behaviour based on tumor size, mitotic activity, cellularity and pleomorphism. It distinguishes SFTs in very low, low, intermediate and high risk tumors. Furthermore, in a series of 110 cases, Demicco et al. proposed a risk stratification model for metastasis and death from disease, identifying three prognostic groups: low, intermediate and high risk patients (68). Nevertheless, no comprehensive studies have been performed to accurately determine the neoplastic aggressiveness of SFT so far. Furthermore, less is known about the genetic ad molecular events responsible for the pathogenesis of SFT, and how this may relate to clinically determined risk factors. In addition, the predicitve value of the few immunohistochemical or molecular biomarkers (including p53, telomerase activity, cycle expression, and Ki67) that have been suggested to have prognostic significance in SFT, remains yet to be clarified.

On gross pathologic examination, SFTs were commonly solid masses, well-circumscribed, encapsulated, and non-infiltrating. Most cases showed prominent vascularity with numerous small and medium size vessels, often adopting focally a hemangiopericytic growth pattern. Typical histological features of SFTs consist in a combination of hypercellular and collagenous areas; in some cases, it can be difficult to evaluate malignancy only according to morphology and immunohistochemical (IHC) features may be useful like positivity of CD34, bcl-2 and STAT6 which are the most important markers for diagnosis, especially in differentiating SFTs from gastrointestinal stromal tumors (GISTS) (53). In particular, STAT6 has recently emerged as a sensitive and specific marker, which identifies the NAB2-STAT6 fusion product (69). Occasionally, SFTs are positive for epithelial membrane antigen (EMA), smooth muscle actin (SMA), and negative for cytokeratin, S100, and desmin. In our review, positivity for vimentin was found in all the 9 cases reporting on IHC (table 1). CD34 and bcl-2 were the immunostainings most frequently used (30 and 11 cases, respectively); the former was positive in 100%, and the latter in 91% of the examined cases (table 1). Frequent immunostaining for CD 99 was detected (90%), as well as negativity for desmin (10 of the 11 reported cases) and smooth muscle actin (SMA).

The ideal therapy for malignant SFTs is uncertain, and the assessment of the most effective clinical management remains to be established. Information about the employed treatments and the prognosis was available in 57 cases. In all these cases surgery was the treatment of choice, with an associated complete omentectomy perfomed in nine (16%) cases. Three cases required an extended resection, with the exeresis of bilateral adnexa (two cases), as well as uterus, appendix and perirectal metastases. Considering the favourable oncological outcomes of SFT, radiotherapy is generally not indicated when surgical excision has resulted in negative margins, and it is usually reserved in cases with incomplete resection or recurrence. Chemotherapy or targeted therapies with tyrosin kinase inhibitors (TKIs) may be recommended especially in unresectable cases, since half of these tumours is positive for cKIT TKIs (70). However, it is difficult to identify the most effective chemotherapy regimen for advanced SFTs on the basis of the current evidence. Cytotoxic doxorubicin based, gemcitabine based, and paclitaxel based schemes are used for patients with locally advanced, recurrent, or metastatic disease, but results have been variable, often with low response rates (71). Several novel targeted strategies, like temozolomide bevacizumab combination therapy, sunitinib, sorafenib and pazopanib have recently shown promising results (72), but further studies are needed to validate their routine use.

Also the prognosis of patients affected by omental

SFT remains difficult to establish. In general, most of the patients with extrapleural disease show a benign clinical course after complete surgical resection. The overall median 5- and 10-year survival rates after surgical resection are 59% to 100% and 40% to 89%, respectively (73). Nevertheless, some omental SFTs showed aggressive malignant behavior resulting in recurrence or metastasis, even many years after the surgical removal. Ten (18%) patients presented distant metastases with a median time between the start of frontline treatment and first recurrence of 25 months (range 3–52). Globally, six (10%) patients died because of the disease, three (5%) were alive with disease, and thirty (53%) were free of disease in the cases reviewed.

CONCLUSIONS

In conclusion, we came across a rare case of giant SFT originating from greater omentum. Imaging, especially CT or MRI, are useful for the detection and anatomic evaluation of these tumors, but only surgery with subsequent pathological examination and immunohistochemical tests can provide a certain diagnosis of SFT. Surgery, with or without radiation and chemotherapy, should be considered for treatment on the basis of the dimensions and extension of the lesions, with a long-term follow-up in order to early detect local recurrences and distant metastases.

Conflict of interest

The authors declare that there are no conflicts of interest.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-chief of tis journal on request.

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