

## Case Report

# Plexiform fibromyxoma of the stomach: a case report and review of the literature

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**Abstract:** Background: Plexiform fibromyxoma (PF) is a recently described rare type of mesenchymal tumor of the stomach that is characterized by a peculiar plexiform growth pattern, myxoid stroma, and spindle-shaped myofibroblastic cells. PFs are mainly located in the antrum of the stomach, and are treated by distal or partial gastrectomy. This study reports a rare case of PF that occurred in the body of the stomach. A review of PF cases reported in both English and Chinese literature is also provided. Case report: A 52-year-old Chinese female presented with 1-month of intermittent upper abdominal pain. Gastroscopy and computed tomography (CT) revealed a submucosal mass with a small overlying mucosal ulceration in the gastric wall at the mid greater curvature of the stomach. Intraoperative rapid frozen section suggested the benign nature of the tumor and the patient underwent gastroscope-assisted laparoscopic wedge resection. Pathological examination of the resected specimen revealed plexiform growth of spindle cells without nuclear atypia and abundant myxoid capillary-rich extracellular matrix. The spindle-shaped cells diffusely expressed smooth muscle actin (SMA) and focally expressed CD10, but were negative for CD34, CD117, DOG-1, P53, and KI-67 (index 1%). The 10-month postoperative course was favorable without any complications or disease recurrence. Conclusion: Although PF closely resembles gastrointestinal stromal tumors (GIST) and other myxoid/fibromyxoid tumors of the stomach, it has distinct pathological and/or immunochemical features, as well as clinical management. Further clinical research is needed to elucidate the clinical course and to develop a standard treatment strategy.

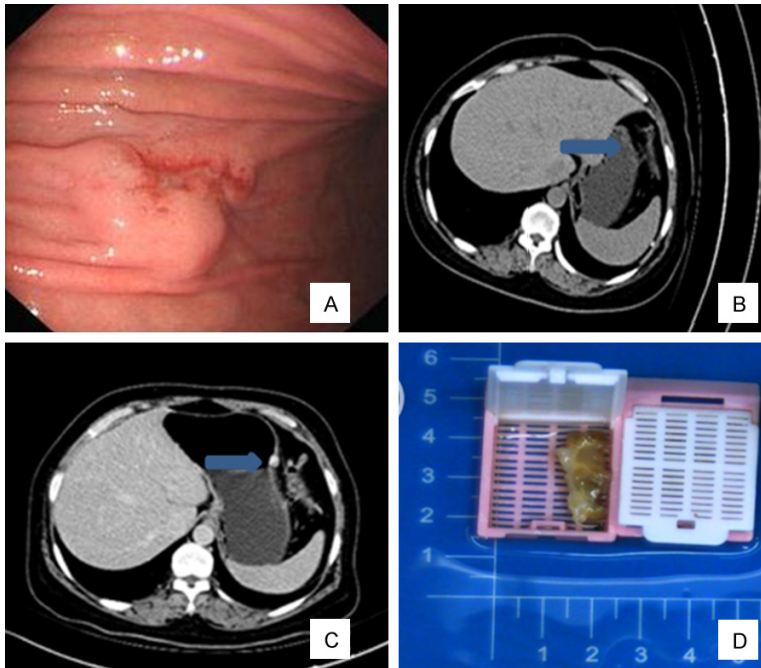
**Keywords:** Plexiform fibromyxoma, plexiform angiomyxoid myofibroblastic tumor, mesenchymal tumor

## Introduction

Gastric plexiform fibromyxoma (PF) is a rare type of mesenchymal tumor with a peculiar plexiform growth pattern, myxoid stroma, and spindle-shaped myofibroblastic cells [1]. The name of this entity has been controversial since Takahashi et al. first reported this unique gastric mesenchymal tumor as "plexiform angiomyxoid myofibroblastic tumor (PAMT)" in 2007 [2]. In 2008, Yoshida et al. described two similar cases as "plexiform angiomyxoid tumor" [3]. Miettinen et al. also reported several cases of similar tumors named "plexiform fibromyxoma" in 2009 [1]. In 2010, this unique gastric mesenchymal tumor was officially designated as "plexiform fibromyxoma" by the World Health Organization (WHO) [4].

To date, approximately 40 cases of gastric PF have been reported in the English literature [5]. This rare type of tumor occurs primarily in adults with an age range from 4 to 75 years and equal gender distribution [6]. PF mostly occurs in the gastric antrum, and the clinical symptoms include upper gastrointestinal bleeding, abdominal distension, abdominal pain, nausea, hematemesis, gastric ulcers, anemia, weight loss or obstruction [7, 8]. Currently, the diagnosis of PF is mainly based on postoperative pathological examination, and sometimes is also confirmed by immunohistochemical tests [1].

In this study, we report a rare case of PF of the stomach, which was removed by laparoscopic wedge resection. Further, we discuss the clini-



**Figure 1.** Preoperative examination of the tumor. (A) Endoscopic image of the tumor showing a submucosal mass measuring 1.0 cm × 1.5 cm with small mucosal ulceration located in the body of the stomach. (B, C) Computed tomography scan of the abdominal demonstrating a heterogeneously-enhancing mass in the gastric wall at the mid greater curvature of the stomach. (D) The resected grey/white-colored tumor measuring 1.0 cm × 1.0 cm × 1.5 cm.

copathological features of this rare entity and review the reported cases in the literature.

### Case presentation

A 52-year-old Chinese female presented to the General Hospital of Ningxia Medical University (Yinchuan, China) with 1-month of intermittent upper abdominal pain in January 2016. Physical examination revealed an upper abdominal pain below the xiphoid process without rebound tenderness. The patient had normal blood test results and was negative for tumor markers CEA, CA125, and CA19-9. Gastroscopy revealed a 1.0 cm × 1.0 cm × 1.5 cm gastric submucosal mass with small overlying ulcerated mucosa (**Figure 1A**). Computed tomography (CT) of the stomach demonstrated a tumor-like lesion in the gastric wall at the mid greater curvature of the stomach (**Figure 1B** and **1C**). The patient underwent gastroscope-assisted laparoscopic wedge resection. Intraoperative rapid frozen section examination suggested the benign nature of the tumor, and therefore extended radical surgery and lymph node dissection was not performed. The resect-

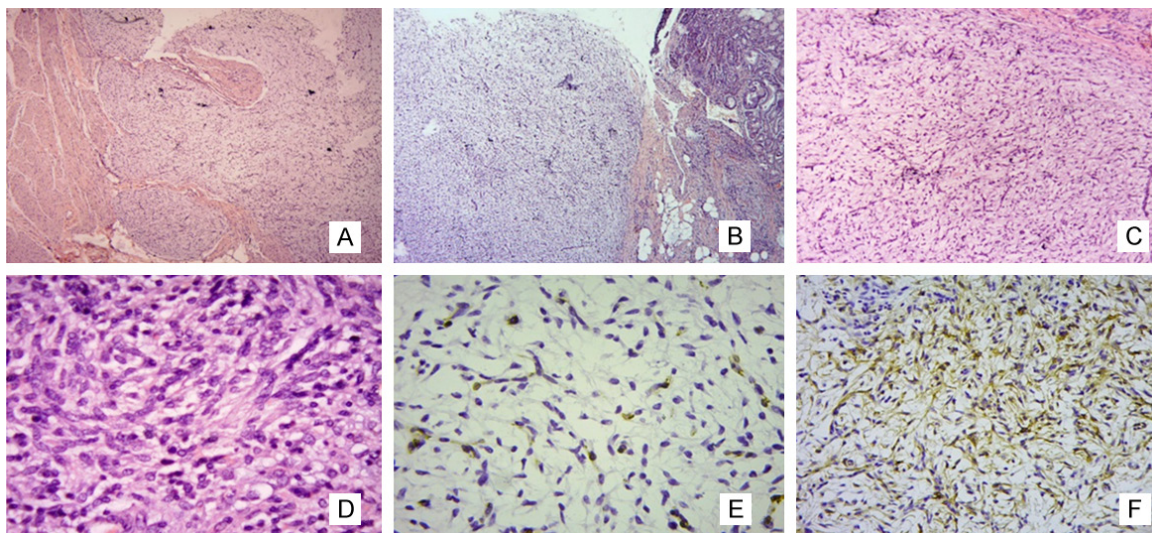
ed specimen appeared as a mucous, white-grey-colored solid mass, measuring 1.0 cm × 1.0 cm × 1.5 cm (**Figure 1D**), located between the submucosa and muscle wall. As shown in **Figure 2A-D**, histopathological examination revealed myxo collagenous nodules and networks of capillaries. The tumor exhibited plexiform or nodular growth of spindle cells without nuclear atypia or mitosis. Abundant myxoid capillary-rich extracellular matrix was observed. Immunohistochemically, the spindle-shaped cells diffusely expressed smooth muscle actin (SMA) (**Figure 2E**) and focally expressed CD10 (**Figure 2F**). In addition, the tumor cells were positive for the epithelial marker cytokeratin protein (CKP) and S100, but were negative for the vascular markers CD34, CD117, DOG-1, p53, desmin, epithelial membrane antigen (EMA),

and KI-67 (index 1%). The postoperative course was favorable without any complications. At 10 months after the operation, no evidence of residual or recurring tumor was detected.

### Discussion

Gastric plexiform fibromyxoma is a rare type of mesenchymal tumor. To date, less than 50 cases have been previously reported in both English and Chinese literature. **Table 1** summarizes a total of 45 documented cases of PF including the one reported in the current study [1, 2, 5, 7, 9-31]. Among the 45 cases, 18 were male patients and 26 were females with a median age of 47 (range: 7-75 years). Most commonly the presenting symptoms include abdominal discomfort and pain, anemia, and upper gastrointestinal bleeding. Other symptoms are also reported, such as weight loss, ulcer, nausea, chest pain, melena, and even hematemesis. PF has occurred in the antrum of the stomach in 88.6% (39/44) of reported cases. The pylorus and/or duodenum are involved in 7 cases [1, 27]. In addition, PF has occasionally been found in the gastric fundus

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**Figure 2.** Pathological and immunohistochemical evaluation of PF. (A, B) Microscopic evaluation (40 ×, hematoxylin & eosin) of PF showing plexiform or nodular growth pattern. (C) Abundant myxoid capillary-rich extracellular matrix was observed (HE 400 ×). (D) The mass contains spindle-to oval-shaped cells without nuclear atypia. Immunohistochemically, the spindle tumor cells diffusely expressed (E) smooth muscle actin and focally expressed (F) CD10 (EnVision method).

[14], pyloroduodenal junction [19] and posterior mediastinum [19]. In this study, PF occurred in the body of the stomach. These findings suggest that PF is not restricted to the stomach although it primarily occurs in the antrum. Immunohistochemically, PFs are positive for SMA in all cases, and for CD10 (11/44, 25.0%), vimentin (8/44, 18.2%) and MSA (8/44, 18.2%) in some cases. The immunoreactivity for SMA suggests that the tumor cells are myofibroblastic in origin with smooth muscle differentiation. Immunoreactivity for HHF35 [10, 18], desmin [7, 17, 21] and caldesmon [19, 21] have also been reported. In addition, the Ki-67 index is generally quite low ( $\leq 1\%$ ), indicating the low level of cell growth in PFs. Typically, PFs do not express CD34, CD117, DOG-1, S-100, CD100,  $\beta$ -catenin, claudin-1, melan A, activin receptor-like kinase 1 (ALK-1), or keratins.

Preoperative diagnosis of PF is often difficult because the disease has nonspecific clinical manifestations and imaging characteristics. Currently, the diagnosis of PF is primarily based on pathological and immunochemical examinations. PF is often misdiagnosed as gastrointestinal stromal tumors (GIST), the most common mesenchymal tumor of the stomach with an estimated incidence rate 150 times higher compared with PF [1], due to common clinical manifestations, imaging characteristics, and

myofibroblastic immunophenotype of the two entities [32]. While PF is characterized by spindle cells, plexiform growth pattern, myxoid matrix, and capillaries, GIST does not show the intramural plexiform growth. Immunohistochemically, all PF cases are typically positive for SMA and negative for CD117, CD34, and DOG-1 [15], whereas GISTs generally express CD117 and DOG-1 (>90% of cases) and CD34 (>80% of cases) [33]. Thus, GISTs can be distinguished from PFs based on both morphological and immunohistochemical findings. Moreover, the differential diagnosis of PFs also includes other myxoid and fibromyxoid tumors of the stomach, including inflammatory fibroid polyp, myxoid leiomyoma, gastric fibromatosis, mesenteric inflammatory myofibroblastic tumor, gastrointestinal schwannoma, and plexiform neurofibroma. Inflammatory fibroid polyps are typically composed of spindled fibroblasts and inflammatory cells, and are positive for CD34 and negative for SMA [34]. Myxoid leiomyoma is composed of relatively large cells with blunt-ended nuclei and fibrillary cytoplasm, and mostly expresses SMA, desmin, and caldesmon. Fibromatosis is composed of evenly distributed large cells and is generally positive for SMA and nuclear  $\beta$ -catenin [35]. Cells of mesenteric inflammatory myofibroblastic tumors demonstrate multi-patterned growth and lymphoplasmacytic infiltration, and are positive for

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**Table 1.** Clinicopathological data for 44 cases of plexiform fibromyxoma reported in both English and Chinese literature

Author, year	No. of cases	Patient Gender/age (years)	Symptoms	Tumor location	Surgery	Immunohistochemistry	Outcome, follow-up duration
Takahashi et al., 2007 [2]	2	M/50	Perforated stomach	Antrum	Distal gastrectomy	SMA(+), MSA(+), CD34(-), S-100(-), c-kit(-)	AWDR, 12 months
		M/68	No significant symptom	Antrum	Distal gastrectomy	SMA(+), MSA(+), CD34(-), S-100(-), c-kit(-)	AWDR, 12 months
Galant et al., 2008 [9]	1	M/61	Hematemesis	Antrum	Distal gastrectomy	SMA(+), Vim(+), CD34(-), CD117(-), S-100(-)	AWDR, 6 months
Rau et al., 2008 [10]	1	F/50	Nausea	Antrum	Wedge resection	SMA(+), HHF35(+), CD34(-), S-100(-), c-kit(-)	AWDR, 3 months
Miettinen et al., 2009 [1]	12	5 M and 7 F/7-75	3 upper gastrointestinal bleeding, 3 ulcer, 2 weight loss, 2 anemia and 2 without significant symptoms	6 Antrum, and 6 antrum + pylorus and/or duodenum	10 partial gastrectomy, 1 subtotal gastrectomy, and 1 gastric wall resection	SMA(+), variable CD10 c-kit(-), DOG-1(-), CD34(-), desmin(-), S100(-)	4 AWDR, 2 alive with unknown disease condition, 3 died of unknown causes, 3 loss to follow up, 2-306 months
Pailoor et al., 2009 [11]	1	F/23	Melena	Antrum	Partial gastrectomy	SMA(+), Vim(+), CD34(-), CD100(-), c-kit(-)	AWDR, 2 months
Sing et al., 2010 [12]	1	F/35	No significant symptom	Antrum	Wide local excision	SMA(+), MSA(+), calponin(+), CD34(-), CD117(-), S-100(-)	AWDR, 12 months
Tan et al., 2010 [13]	1	M/60	Abdominal discomfort and mass	Antrum	Distal gastrectomy	SMA(+), MSA(+), CD34(-), CD100(-), c-kit(-)	N/A
Wang et al., 2010 [14]	1	F/54	Abdominal distension and loss of appetite	Fundus	Endoscopic resection	SMA(+), Vim(+), CD34(-), CD117(-), S-100(-)	AWDR, 6 months
Kim et al., 2011 [15]	1	M/52	Dyspepsia	Antrum	Wedge resection	SMA(+), CD34(-), CD117(-), S-100(-)	AWDR, 5 months
Cai et al., 2012[16]	2	M/32	No significant symptom	Antrum	Distal gastrectomy	SMA(+), MSA(+), CD34(-), CD117(-), S-100(-)	N/A
		F/47	Upper abdominal pain	Antrum	Radical distal gastrectomy	SMA(+), MSA(+), CD34(-), CD117(-), S-100(-)	N/A
Kang et al., 2012 [17]	2	M/47	Gastric mass	Antrum	Wedge resection	SMA(+), CD10(+), c-kit(-), CD34(-), ER(-), PgR(-) and claudin-1(-) MIB-1(+), desmin(+) in only small number of tumor cells.	Loss to follow up
		F/63	No significant symptom	Antrum	Endoscopic resection	SMA(+), CD10(+), c-kit(-), CD34(-), ER(-), PgR(-) and claudin-1(-)	Loss to follow up
Wang et al., 2012 [18]	1	M/12	Gastrointestinal bleeding	Antrum	Partial gastrectomy	SMA(+), HHF35(+), CD34(-), CD117(-), S-100(-), DOG-1(-)	AWDR, 84 months
Duckworth et al., 2014 [19]	2	F/11	Anemia	Pyloroduodenal junction	Resection	SMA(+), S-100(-), CD34(-), desmin(-), CD117(-)	AWDR, 15 months
		F/16	Chest pain	Posterior mediastinum	Distal gastrectomy	SMA(+), S-100(-), CD34(-), desmin(-), CD117(-)	AWDR, 14 months
Ikemura et al., 2014 [7]	1	F/27	Abdominal pain, melena	Antrum	Partial gastrectomy	SMA(+), CD10(+) c-kit(-), CD34(-), ER(-), PgR(-), claudin-1(-) desmin(+), MIB-1(+) in only a small portion of tumor cells	AWDR, 40 months

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Lee et al., 2014 [20]	1	F/42	Abdominal pain, fever, anemia	Antrum	Distal gastrectomy	SMA(+) desmin(-), CD34(-) and S100(-) Ki-67(index 40%)	AWDR, less than 1 month
Li et al., 2014 [21]	1	F/32	No significant symptoms	Antrum	Partial gastrectomy	SMA(+), Vim(+), caldesmon(+), desmin(+) CD117(-), CD34(-), DOG-1(-), S-100(-), ALK(-), $\beta$ -catenin(-), Ki-67(index 1%)	AWDR, 36 months
Li et al., 2014 [22]	1	F/73	Upper abdominal pain	Antrum	Partial gastrectomy	SMA(+), Vim(+), Bcl-2(+) CD34(-), CD117(-), S-100(-)	N/A
Tian et al., 2014 [23]	1	M/64	Upper abdominal discomfort	Antrum	Distal gastrectomy	SMA(+), CD34(-), CD100(-), CD117(-), DOG-1(-)	AWDR, 6 months
Ni et al., 2015 [24]	1	F/21	Gastrointestinal bleeding	Antrum	Distal gastrectomy	SMA(+), Vim(+), CD34(-), CD117(-), S-100(-), EMA(-)	AWDR, 36 months
Wei et al., 2015 [25]	1	F/50	Upper abdominal pain	Antrum	Subtotal gastrectomy	SMA(+), CD100(-), S-100(-), DOG-1(-), Bcl-2(-)	AWDR, 3 months
Yue et al., 2015 [26]	2	M/34	Upper abdominal pain	Antrum	Distal gastrectomy	SMA(+), MSA(+) CD34(-), CD117(-), S-100(-), DOG-1(-)	AWDR, 24 months
		F/50	Upper abdominal pain	Antrum	Subtotal gastrectomy	SMA(+), MSA(+) CD34(-), CD117(-), S-100(-), DOG-1(-)	AWDR, 3 months
Dixit et al., 2016 [27]	1	F/51	Intermittent abdominal pain, vomiting and weight loss	Pylorus	Distal gastrectomy	SMA(+) CD117(-), CD34(-), S-100(-), ALK(-), $\beta$ -catenin(-), caldesmon(-)	N/A
Jonaitis et al., 2016 [31]	1	F/28	Weight loss, vomiting, nausea and upper abdominal discomfort	Antrum	Partial gastrectomy	SMA(+) desmin(-), CD34(-), S100(-), Ki-67(index 40%)	N/A
Kane et al., 2016 [5]	1	F/28	Abdominal pain, anemia	Antrum	Distal gastrectomy	SMA(+), CD10(+) CD117(-), DOG-1(-), CD34(-), S-100(-), D2-40(-), keratin AE1/3(-), $\beta$ -catenin(-)	AWDR, 23 months
Li et al., 2016 [29]	1	M/11	Upper abdominal discomfort	Antrum	Partial gastrectomy	SMA(+), caldesmon(+) CD34(-), CD100(-), CD117(-)	AWDR, 12 months
Quero et al., 2016 [30]	1	M/47	Regurgitation and epigastric discomfort	Antrum	Distal gastrectomy	Vim(+), a-SMA(+), DES(partially+), CAL(partially+), AE1/AE3(focal+), panCK(focal+), CD10(partially+) CD117(-), DOG-1(-), S100(-), SYN(-), CHR(-), ALK(-), CD34(-), CAM5.2(-), CK20(-), CK7(-), EMA(-), CDX2(-), P53(-), melanA(-), HMB45(-)	AWDR, 10 months
Zhang et al., 2016 [31]	1	M/48	Upper abdominal pain	Antrum	Partial gastrectomy	SMA(+), Vim(+) CD34(-), CD117(-), S-100(-), DOG-1(-)	AWDR, 12 months
Present case, 2016	1	F/52	Intermittent upper abdominal pain	The body of stomach	Wedge resection	SMA(+), CD10(+), S-100(+), CK-P(+), CD34(-) P53(-), CD117(-), DOG-1(-), desmin(-), EMA(-), Ki-67(index 1%)	AWDR, 10 months

F, female; M, male; SMA, smooth muscle actin; MSA: muscle specific actin; VIM, vimentin; AWDR: alive without disease recurrence; ALK, anaplastic lymphoma kinase; EMA: epithelial membrane antigen; and N/A, not applicable.

SMA and ALK-1 in more than 40% of cases [36]. Gastrointestinal schwannomas are positive for S-100, and negative for SMA [37]. Plexiform neurofibroma exhibits immunoreactivity for S-100 [38].

PFs exhibit no significant nuclear atypia or mitotic activity, and thus is generally considered a benign tumor. Among the 44 documented cases in the literature, 28 patients have been alive without disease recurrence or metastasis during follow-up periods ranging from 1 to 306 months (median: 12 months). Consistently, Takahashi reported 15 cases of PF, among which no recurrence had occurred during a follow-up of 2 months to 25.5 years after surgery (median: 3 years). Nevertheless, the possibility of malignancy can not be completely ruled out in some PF cases. For instance, PFs with extra-gastric extension and/or lymphatic/vascular invasion are occasionally reported [10]. A PF case showing vascular invasion, necrosis and bleeding has also been documented [33]. Long-term follow-up is highly recommended to monitor the condition of such cases.

PFs mainly occur in the gastric antrum, and therefore are usually resected by partial or distal gastrectomy. As shown in **Table 1**, 31 out of the 44 reported cases underwent partial or distal gastrectomy. Local wedge resection was performed in a small number of cases (4/44, 9.1%), including the patient in the current study. In a PF case reported by Tian et al., the patient was initially treated with endoscopic submucosal dissection (ESD), but was given distal gastrectomy due to heavy bleeding after the ESD [23]. Another PF case was confirmed as low-grade malignant tumors via intraoperative rapid pathological diagnosis and was then removed by radical distal gastrectomy [16]. Dissection without precise detection of the tumor range may result in either incomplete resection of the tumor or excessive resection of the gastric wall, leading to disease recurrence or deformation of the stomach. Therefore, accurate determination of surgical margin is required to ensure the prognosis in PF patients.

In summary, we herein have reported a case of PF, a rare mesenchymal gastric tumor that requires differential diagnosis from GIST and other mesenchymal tumors of the stomach.

Pathological and immunohistochemical features of PF may aid in separating the tumor from other entities. Partial or distal gastrectomy is the primary surgical treatment for PF. Although the short-term prognosis has been good with no recurrence or metastasis, the true biologic nature of PF remains unknown largely due to its rarity and the limited follow-up period. Further clinical research is necessary to elucidate the clinical course and to develop a standard treatment strategy.

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The reported patient signed written informed consent for the publication of images and medical data in this case report.

### Disclosure of conflict of interest

None.

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