# Case Report Myopericytoma of the stomach: report of one case and review of literature

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**Abstract:** Myopericytoma is an uncommon, slow-growing benign tumour of concentrically distributed perivascular myoid cells, that occurs generally in the skin and superficial soft tissues especially in distal extremities. In the visceral organs, it is particularly rare. We provide the first report of this rare entity in the stomach. A 45-year-male presented to an outside hospital because of pharyngalgia and cough 10 days prior. Endoscopic ultrasonography revealed a 0.92 cm × 0.92 cm hypoechoic lesion in the submucosa of sinuses ventriculi. For further diagnosis and treatment, the patient came to our hospital, and underwent endoscopic submucosal excavation (ESE), without adjuvant therapy. Postoperative pathology was myopericytoma. No recurrence was found in the follow-up of 27 months. In conclusion, myopericytoma is a comparatively newly described disease entity approved by the World Health Organization classification for tumours of soft tissue. The present report shows the first case of myopericytoma of the stomachto remind clinicians and pathologists that myopericytoma may be encountered at this location.

Keywords: Myopericytoma, stomach, histopathology

#### Introduction

Myopericytoma is a benign tumour of concentrically distributed perivascular myoid cells. Myopericytoma, including myofibroma, is classified under the broad category of pericytic neoplasms in the 2013 WHO classification for tumours of soft tissue, which also includes angioleiomyoma and glomus tumor [1]. Histologically, myopericytoma has a concentric proliferation of oval- to spindle-shaped cells with a perivascular arrangement [2-4]. These cells express muscle-specific actin and alpha-smooth muscle actin (ASMA), whereas desmin is expressed rarely, so they are regarded as modified smooth muscle cells [5]. The neoplasm generally arises from the skin and superficial soft tissues of distal extremities, and is particularly rare in visceral organs [2] and had hitherto not been reported in the stomach. Here, we present a man who was diagnosed with myopericytoma of the stomach, aiming to raise awareness about this entity and the need to differentiate it from usual carcinoma of the stomach or gastrointestinal stromal tumor (GIST).

#### Case report

A 45-year-male patient presented to an outside hospital with pharyngalgia and cough 10 days prior, mainly with irritating dry cough, but no expectoration, dyspnea, hemoptysis, fever, acid reflux, hiccups, nausea and vomiting, nor bellyache and diarrhea. Endoscopic ultrasonography revealed a 0.92 cm × 0.92 cm hypoechoic lesion in the submucosa of sinuses ventriculi. For further diagnosis and treatment, the patient came to our hospital. He denied any positive family history or history of past illness. The physical examination showed no apparent abnormality. All laboratory data were within normal limits. Gastroscopy showed a 1.5 cm × 1.5 cm lesion in the submucosa of sinuses ventriculi close to the corpora ventriculi (Figure 1A). The patient underwent endoscopic submucosal excavation (ESE), without adjuvant therapy (Figure 1B). The pathologic diag-



**Figure 1.** Gastroscopy showed a 1.5 cm × 1.5 cm lesion in the submucosa of sinuses ventriculi close to corpora ventriculi (A). The patient underwent endoscopic submucosal excavation (ESE) (B).

nosis was myopericytoma. The patient was free of disease after follow-up for 27 months.

## Materials and methods

## Immunohistochemical staining

Muscle tissue was used as a negative control, and tonsil tissue was taken as a positive control. Serial sections at 4  $\mu$ m were taken on poly-lysine slides and baked at 60°C in the oven for 2 h. Slides were soaked in xylene I and xylene II for 15 min and washed for 5 min

in anhydrous, 95%, 85%, and 70% gradient alcohol, respectively, and then washed with PBS 3 times for 2 min. The tissue antigen was retrieved according to the requirements of each antibody (e.g., citric acid high pressure for 1.5 min). One drop or 50  $\mu$ L of 3% hydrogen peroxide was added to each section and incubated at room temperature for 10 minutes to block endogenous peroxidase. 1 drop or 50  $\mu$ L of non-immune goat serum was added to each section and incubated for 10 minutes at room temperature. 50  $\mu$ L of the primary antibody was added to each section and incubated.



**Figure 2.** The tumor cells were arranged in nests or fascicles (A) (original magnification  $\times$  50). The neoplasm was characterized by numerous small variably sized, thin-walled, branching blood vessels (B) (original magnification  $\times$  100). The oval to spindle cells possessed indistinct cell borders, and lightly eosinophilic cytoplasm (C) (original magnification  $\times$  200). Nuclear atypia, mitotic figures, and necrosis were not observed (D) (original magnification  $\times$  400). The neoplastic cells exhibited immunoreactivity for smooth muscle actin (E) (original magnification  $\times$  200). h-Caldesmon stained the neoplastic cells (F) (original magnification  $\times$  100).

ed for 60 minutes at room temperature. 50  $\mu$ L of the ready-to-use MaxVision<sup>TM</sup> (KIT-5910 MaxVision<sup>TM</sup> HRP-Polymer anti-rabbit/mouse IHC Kit) reagent was added and incubated for 15 minutes at room temperature. 100  $\mu$ L of freshly prepared DAB coloring solution for 5 minutes was added and slides were observed under a microscope. Slides were rinsed with water and counterstained with hematoxylin, gradient-dehydrated with alcohol and hyalinized with xylene, and then sealed with neutral gum.

# **Pathologic features**

#### Gross features

Dissection revealed a non-encapsulated, grayish-white solid tumor measuring  $1.5 \times 1.0 \times 0.6$  cm, pale tan cut surface with no evidence of hemorrhage or necrosis.

## Microscopic features

Tumor was non-encapsulated, well-circumscribed vascular neoplasm, with proliferating bland ovoid to spindle cells with eosinophilic cytoplasm. The tumour cells were arranged in nests, nodositas or in a concentric perivascular form, surrounded by a large number of thin-walled, variably sized, branching blood vessels (Figure 2A-C). Nuclear atypia, mitotic figures, and necrosis were not observed (Figure 2D). The tumour cells were positive for vimentin, smooth muscle actin (Figure 2E) and h-Caldesmon (Figure **2F**), and had a Ki-67 index of < 5%. Tumor demonstrated a vascular proliferative pattern with CD34 antibody which stained endothelial cells. However, staining was negative for desmin, cytokeratin, CD117, DOG1, S-100 protein, human melanoma black (HMB)-45, synaptophysin, and CD56.

## Review of literature on visceral myopericytoma

After an extensive search of the English literature in Pubmed, we found 23 cases of visceral myopericytoma. The brief clinical information of these cases is summarized in Table 1. Among the 23 cases, 10 were male and 13 were female, with a median age of 52 years (range, 26 to 75 y). Visceral myopericytomas most often (10/23, 43.5%) occurred in the kidney [2, 6-9], followed by the lung (4/23,17.4%) [10-13], 2 cases (2/23, 8.7%) of bladder [7, 14] and 2 cases (2/23, 8.7%) of uterus [15]; the other 5 cases occurred in the heart [16], liver [17], periampullary [18], ovary [15], and the space between liver and stomach (1/23, 4.3%) [19]. Of the 23 cases reported, only one in the ampulla of the duodenum was malignant, which was associated with Epstein Barr virus infection. Interestingly, myopericytoma occurring in the lung is characterized by multiple nodules in both lungs. All patients were treated by simple operation, and all of the patients evaluated (21/21, 100.0%) had no recurrence after the surgery during the followup period (median follow-up, 26 mo; range, 5 to 86 mo).

Case#	References	Age (y)	Sex	Size (cm)	Location	Symptoms	Treatment	Outcome (mo)
1[6]	Jun Li et al.	56	F	1.8	Kidney	Pain on the right side of the abdomen	Surgery	N-Re, 66
2 [6]	Jun Li et al.	33	Μ	4.5	Kidney	Asymptomatic	Surgery	N-Re, 64
3 [6]	Jun Li et al.	46	Μ	7.3	Kidney	Asymptomatic	Surgery	N-Re, 46
4 [6]	Jun Li et al.	70	Μ	4.8	Kidney	Asymptomatic	Surgery	N-Re, 16
5 [6]	Jun Li et al.	69	Μ	4.2	Kidney	Asymptomatic	Surgery	N-Re, 14
6 [6]	Jun Li et al.	59	Μ	3.6	Kidney	Asymptomatic	Surgery	N-Re, 26
7 [9]	Sadhna Dhingra et al.	40	F	3.8	Kidney	Pain on the left side of the abdomen and frequent urination	Surgery	N-Re, 24
8 [8]	Zhiqiang Zhang et al.	39	Μ	18	Kidney	Asymptomatic	Surgery	N-Re, 20
9 [7]	Ming Zhao et al.	59	F	3.6	Kidney	Urinary frequency and dysuria	Surgery	N-Re, 14
10 [2]	Sean K Lau et al.	59	F	3	Kidney	Upper respiratory tract symptoms and hemoptysis	Surgery	N-Re, 8
11 [10]	Jeong Min Mun et al.	63	F	4.5	Lung	Asymptomatic	Surgery	N-Re, 34
12 [11]	Xiao-lian Song et al.	26	Μ	1.7	Lung	Dry cough	Surgery	N-Re, 36
13 [12]	Jian-Hua Cao et al.	52	F	3.5	Lung	Asymptomatic	Surgery	N-Re, 36
14 [13]	Allison Edgecombe et al.	58	F	2.7	Lung	Asymptomatic	Surgery	NA
15 [14]	Takahiro Nagai et al.	75	Μ	2.1	Bladder	Asymptomatic gross hematuria	Surgery	N-Re, 5
16 [7]	Ming Zhao et al.	52	F	6	Bladder	Urinary frequency and dysuria	Surgery	N-Re, 39
17 [15]	Fulvio Borella et al.	49	F	16	Uterus	Progressive pelvic/abdominal pain	Surgery	N-Re, 30
18 [15]	Fulvio Borella et al.	49	F	NA	Uterus	Metrorrhagia	Surgery	N-Re, 86
19 [15]	Fulvio Borella et al.	26	F	NA	Ovary	Pain located in the leftiliac fossa	Surgery	N-Re, 11
20 [17]	Jun Li et al.	62	F	6	Liver	Asymptomatic	Surgery	N-Re, 28
21 [40]	Tomás Francisco Cianciulli et al.	46	F	4.5	Heart	Progressive dyspnea	Surgery	N-Re, 24
22 [19]	Zhihua Chen et al.	51	Μ	15	Liver and stomach space	Abdominal mass and right upper abdominal pain	Surgery	N-Re, 5
23 [18]	Ramdial PK et al.	30	Μ	5	Periampullary	Progressive jaundice	Surgery	Ce, 8

Table 1. Clinical features of the 23 cases of visceral myopericytoma reported in the literatures

Ce, censored; F, female; M, male; mo, months; N-Re, no relapse; NA, not available.

# Discussion

Named by Zimmermann in 1923, pericytes are contractile perivascular cells that enwrap endothelial cells and regulate microvasculature [20]. In 1942, Stout and Murray [21] designated a class of tumours with rounded cells and a presumed pericytic origin as "hemangiopericytoma". Subsequently, the terminology "hemangiopericytoma" was expanded by Enzinger and Smith [22] to include cases with spindled morphology and gaping vascular spaces. The concept of myopericytes was originally proposed by Dictor et al. in 1992 to describe an unusual tumour in a boy. Tumour cells that the authors termed "myopericytes" displayed features intermediate between pericytes and vascular smooth muscle cells [23]. Soon afterwards, the term was used by Requena et al. in 1996 as an alternative name for myofibroma [4]. In 1998, in order to describe such tumours histologically characterized by a striking concentric perivascular proliferation of spindle cells, showing apparent differentiation towards perivascular myoid cells, Granter et al. adopted the term myopericytoma [3]. Myopericytoma, including myofibroma, is classified under the broad category of pericytic neoplasms in the system of 2013 WHO classification for tumours of soft tissue, which also includes angioleiomyoma and glomus tumor [1].

Myopericytoma presents typically as a painless, discrete mass with a wide age range and a male predominance. Anatomic distribution includes most commonly skin or soft tissue in the extremities, followed by head and neck region or trunk [3, 24, 25], and seldom brain and spinal cord [26-29], oral cavity [30], parotid [31], lung [32], kidney [6], urinary tract [7], ampulla of duodenum [18], and intravascular locations [24, 33]. Myopericytoma may be associated with chronic scar or trauma [34, 35]. In rare cases involving viscera, association with Epstein-Barr virus infection in the setting of immunodeficiency has been documented [18, 32]. Myopericytoma is most often solitary; multifocal myopericytomas are rare. Yin and Fletcher made a clinicopathologic analysis of 11 cases of multifocal myopericytomas with molecular identification [36]. To the best of our knowledge, the present case is the first reported gastric myopericytoma.

Histologically, myopericytoma shows a wide range of growth patterns [30]. The histologic features of the tumours are the presence of numerous blood vessels with a concentric perivascular arrangement of ovoid, plump, spindled and/or round myoid cells with eosinophilic cytoplasm and myoid differentiation [2-4]. The neoplastic cells are diffusely immunoreactive for vimentin, smooth muscle actin, and often for h-Caldesmon, whereas desmin is usually negative, which is characteristic of myopericytoma and useful for its differential diagnosis [2-4, 36]. The diagnosis of myopericytoma was made on the basis of the prominent morphologic and immunoreactive findings.

Myopericytoma is generally a painless, slowgrowing neoplasm, rarely measuring > 2 cm, except for a few with infiltrative growth. Malignant forms have also been reported even though extremely uncommon. If the tumor displays cellular pleomorphism, high mitotic activity, or necrosis, it should be diagnosed as malignant myopericytoma, which may lead to local recurrence and rarely metastases [37].

The differential diagnosis includes a series of tumors, such as gastric epithelial neoplasms, gastrointestinal stromal tumours (GIST), and neuroendocrine neoplasms. Simultaneously, other members of the perivascular tumours, including angioleiomyoma, hemangioperithelioma, myofibroma and glomus tumour, should be taken into consideration. In gastric adenocarcinoma, the tumour cells present with remarkable atypia and mitosis, positive staining for epithelial markers (cytokeratins). The immunohistochemical results should be positive for SMA andvimentin, and negative for S-100, CD117, DOG-1. That can help to distinguish this tumour from neurogenic tumours and GIST. The nonexpression of tumour cells of HMB45 and S-100 does not support the diagnosis of malignant melanoma. Finally, neuroendocrine tumours are excluded due to the negative expression of endocrine markers (synaptophysin, CD56). Despite the overlapping of morphologic features with angioleiomyoma, myopericytoma represents a broad morphologic spectrum characterized by distinct concentrically, perivascularly growing myoid tumour cells that stain positively for smooth muscle actin and often for h-Caldesmon, whereas they remain negative for desmin, suggesting a less-differ-

entiated smooth muscle phenotype. Angioleiomyoma is usually a painful nodule occuring commonly on the lower limbs of females. Angioleiomyoma are tumours composed of mature smooth muscle cell bundles with abundant vascular channels that frequently show desmin immunoreactivity in the smooth muscle bundles. Myopericytoma, including myofibroma, is classified under the broad category of pericytic neoplasms in the 2013 WHO classification for tumours of soft tissue, and the latter appears as a subtype of the former. Myopericytoma is arranged in a concentric fashion around vessels with plump spindle cells while the characteristic of myofibroma is mainly expressed as a typical biphasic pattern of central primitive spindle cells and peripheral myoid/myofbroblastic cells [2, 38]. Glomus tumours exhibit a perivascular pattern of growth with cuboidal epithelioid cells, have an organoid pattern of the glomus organ, and lack the characteristic perivascular concentric growth of myopericytoma [2, 24, 38, 39].

Myopericytoma is a newly described tumour approved by the WHO classification. It is an uncommon benign spindle cell tumour, having the unique characteristic of a vascular architectural pattern resembling hemangiopericytoma and showing features of perivascular myoid differentiation. In summary, we report the first case of primary gastric myopericytoma, in order that myopericytoma may be considered as a differential diagnosis when the clinicians and pathologists encounter a similar situation.

In summary, we report the first case of primary gastric myopericytoma. Myopericytoma needs to be considered as a differential diagnosis in clinicopathologic work.

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Written informed consent for publication of his clinical details and clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

## Disclosure of conflict of interest

None.

## Abbreviations

ESE, endoscopic submucosal excavation; WHO, World Health Organization; ASMA, alphasmooth muscle actin; GIST, gastrointestinal stromal tumors.

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