Unusual focal keratin expression in plexiform angiomyxoid myofibroblastic tumor
A case report and review of the literature
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Abstract
Background: Plexiform angiomyxoid myofibroblastic tumor (PAMT), also known as plexiform fibromyxoma, is a rare distinctive benign intramural tumor, typical of gastric antrum, commonly causing mucosal ulceration with upper gastrointestinal bleeding and anemia, effectively treated by complete surgical resection usually accomplished by distal gastrectomy.

Methods and Results: We herein report a 47-year-old man presenting with a syncopal episode, regurgitation and epigastric discomfort, bearing a gastric antral myxoid plexiform tumor positive for α-smooth muscle actin, vimentin and, partially, for caldesmon, desmin, and CD10; CD117, DOG1, CD34, S100, CAM5.2, CK20, CK7, EMA, p53, CDX2, chromogranin A, synaptophysin, anaplastic lymphoma kinase, Melan-A, and HMB-45 were all negative. All these features are typical of PAMT. Of note, focal positivity for AE1/AE3 and pan-CK KL1 was also present.

Conclusions: The finding of a focal keratin expression in PAMT contributes to enlarge the immunophenotypic spectrum of this tumor type and is relevant for avoiding presurgical misdiagnoses which could ultimately lead to inappropriate overtreatment of patients with PAMT.

Abbreviations: CK = cytokeratin, IHC = immunohistochemistry, PAMT = plexiform angiomyxoid myofibroblastic tumor.

Keywords: differential diagnosis, histopathology, immunohistochemistry, plexiform angiomyxoid myofibroblastic tumor, plexiform fibromyxoma

1. Introduction
Plexiform angiomyxoid myofibroblastic tumor (PAMT), also known as plexiform fibromyxoma, is a rare mesenchymal neoplasm, recently characterized by Takahashi et al[1] and Miettinen et al[2] following reports from the preimmunohistochemistry (IHC) era probably concerning the same entity.[3–7] At the best of our knowledge, 59 cases of PAMT (including the present case and 2 uncertain ones) have been reported so far (Table 1).[1],[2,3–33] This tumor affects both sexes with a wide age span (7–75 years). It typically arises in the gastric antrum; exceptional extragastric cases have been described in the esophagus, duodenum, jejunum, gallbladder and, possibly, in the colon.[22,26,27,30,33] PAMT characteristically features a plexiform architecture, with myxoid nodules located in the gastric muscularis propria, often ulcerating the overlying mucosa, composed of ovoid cells with indistinct cytoplasm; a prominent capillary network is invariably present. At IHC, PAMT is positive for α-smooth muscle actin (α-SMA) and, sometimes, for desmin and/or caldesmon, consistently with myofibroblastic differentiation. KIT and PDGFRα are wild type. PAMT pursues a benign course following complete excision by distal gastrectomy. Despite PAMT typical location and plexiform architecture, its rarity and rather vague histology, in a context usually suggestive for GIST (the most frequent gastric mesenchymal tumor[34]), can hinder biopict attempts to achieve a correct preoperative diagnosis. The latter can be further confused by the exceptional feature we herein describe in a PAMT: keratin expression.

2. Case report
A 47-year-old man presented with a syncopal episode following several months of regurgitation and worsening epigastric discomfort. Routine laboratory tests, electrocardiogram, and chest X-ray were unremarkable. Endoscopy showed a subepithelial lesion in the gastric antrum; the overlying mucosa was focally ulcerated. Endoscopic ultrasound–fine needle tissue acquisition[31] did not yield diagnostic material. Contrast-enhanced computed tomography showed an enhancing 6.5 cm mass bulging into the antral cavity and focally involving the omentum. A distal gastrectomy was performed. Currently, at 10 months’ follow-up, the patient is well.

Patient’s informed consent was obtained for publication of this case. All the tests performed were part of the diagnostic work-up,
| Case no. | Author, year | Age, y | Sex | Site | Size, mm | Ulcer | Immunophenotype | KIT/PDGFRA genotype | Treatment | FL, mo |
|---------|--------------|--------|-----|------|----------|-------|-----------------|----------------------|------------|-------|
| 1, 2    | Takahashi et al, 2007 | 50, 68 | M (2) | A (2) | 40, 45 | y, n | VIM+; α-SMA+; HGF+; DES+; CAL+ (focal); CD117+; S100+; NF-; CD34+; OK- | WT (2) | DG, PG | NR, ANED 12 |
| 3       | Rau et al, 2008 | 50 | F | A | 19 | y | α-SMA+; HHF35+; DES+ (focal); CD117+; S100-; NSE-; ALK+; β-Catenin+; CD34- | WT | LE | ANED 3 |
| 4, 5    | Galant et al, 2008 | 61 | M | A | 37 | n | α-SMA+; HHF35+; DES+ (focal); CAL+ (sparsely); CP+ (sparsely); CD117-; PDGFRA-; S100-; Collagen M-; (1); Laminin-; (3); BSE-; (1); CD34-; AE1/AE3-; (0); EMA-; (1); CD10+; ER-; PR-; (1) | NR | PG | ANED 6 |
| 6, 7    | Yoshida et al, 2009 | 19, 46 | F, M | A (2) | 45, 35 | y, n | α-SMA+; HHF35+; DES+ (focal); CAL+ (sparsely); CP+ (sparsely); CD117-; PDGFRA-; S100-; Collagen M-; (1); Laminin-; (3); BSE-; (1); CD34-; AE1/AE3-; (0); EMA-; (1); CD10+; ER-; PR-; (1) | NR | PG | ANED 9 and 4 |
| 8–19    | Palibot et al, 2009 | 23 | F | A (5) | 60 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+ (focal); (1) | NR | PG | ANED 2 |
|          | Metten et al, 2009 | 38, 43, 50, 56, 62, 65, 75 | F | A (5) | 30, 40, 50 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+ (focal); (1) | NR | PG | ANED 2 |
| 20      | Takahashi et al, 2010 | 23 | M | A | 140 | NR | Data compiled with other 18 cases; only the following results, common to all cases, can be attributed with certainty to the single previously unreported case: VIM+; HHF35+; CD117+; S100-; NF-; ALK+; CD34-; OK-; EMA- | NR | PG | ANED 12 |
| 21      | Wang et al, 2010 | 54 | F | GF | 15 | n | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR+; ACTH+; GH+ | NR | ENR | ANED 5 |
| 22      | Sing et al, 2010 | 35 | M | AP | 40 | n | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER+; PR-; (1) | NR | ENR | LE |
| 23      | Tan et al, 2010 | 34 | M | A | 32 (solid + 24% pseudoplastic) | y | α-SMA+; DES+; CD117+; S100-; CD34-; MFH16+ | WT | DG | ANED 2 |
| 24      | Kim et al, 2011 | 52 | M | A | 35 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; EMA- | NR | ENR | ANED 5 |
| 25      | Schul et al, 2012 | 59 | M | P | 15 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; EMA- | NR | ENR | NR |
| 26, 27  | Kang et al, 2012 | 47, 63 | F, M | GB (2) | 30, 22 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; EMA- | NR | ENR | ANED 72 |
| 28–30   | Bi et al, 2012 | 31, 42, 47 | F (2), M (3) | A (3) | 45, 66, 48 | y | VIM+; α-SMA+; HHF35+; CAL+; CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; EMA- | NR | ENR | NR |
| 31      | Li et al, 2014 | 32 | M | A | 34 | n | VIM+; α-SMA+ (partial); DES+ (partial); CAL+ (partial); CD117+; D2-40+; S100-; ALK-; β-Catenin-; CD34- | NR | PG | ANED 36 |
| 32, 33  | Duckworth et al, 2014 | 11, 16 | F (2) | E, PD | 32, 35 | y | VIM+; α-SMA+ (focal); CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; EMA- | NR | ENR | ANED 14 and 15 |
| 34      | Ikemura et al, 2014 | 27 | F | A | 30 | y | VIM+; α-SMA+ (focal); CD117+; S100-; CD34+; AE1/AE3-; CD10+; ER-; PR-; CD99+ | NR | PG | ANED 40 |
| 35      | Lee et al, 2014 | 42 | F | A | 129 | y | α-SMA+; HHF35+; DES+; CD117+; S100-; ALK-; β-Catenin-; CD34-; MFH16+; CD117-; D2-40+; CD99+; CD34-; MUC4+ | NR | DG | ANED 1 |
| 36      | Sakamoto et al, 2014 | 60 | M | A | 20 | n | α-SMA+; CD117+; S100-; CD34+; CD10+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | NR | PG | ANED 12 |
| 37      | Banerjee et al, 2015 | 19 | F | D | 138 | n | VIM+; α-SMA+; HHF35+; DES+; CP+; CD117+; S100-; CD34+; CD10+; ER-; HMFG4+; CD34+; CD99+; smooth muscle+; Ki-67+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | NR | DG + POI | ANED 6 |
| 38      | Lu et al, 2015 | 26 | F | A | NR | n | VIM+; α-SMA+; HHF35+; DES+; CD117+; S100-; CD34+; CD10+; ER-; HMFG4+; CD34+; CD99+; smooth muscle+; Ki-67+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | NR | DG | NR |
| 39      | Passam et al, 2015 | 55 | F | G | 10 | n | VIM+; α-SMA+; HHF35+; DES+; CD117+; S100-; CD34+; CD10+; ER-; HMFG4+; CD34+; CD99+; smooth muscle+; Ki-67+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | WT | C | NR |
| 40      | Moritz et al, 2016 | 9 | F | A | 40 | n | VIM+; α-SMA+; HHF35+; DES+; CD117+; S100-; CD34+; CD10+; ER-; HMFG4+; CD34+; CD99+; smooth muscle+; Ki-67+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | NR | P | ANED 4 |
| 41      | Kane et al, 2016 | 28 | F | A | 55 | n | VIM+; α-SMA+; HHF35+; DES+; CD117+; S100-; CD34+; CD10+; ER-; HMFG4+; CD34+; CD99+; smooth muscle+; Ki-67+; CD99+; CD34+; MUC4+; DOG1+; NSE+; GFAP+; keratin+; epithelial+; smooth muscle+; Ki-67+ | NR | DG | ANED 23 |
and followed standard laboratory procedures. This case is not part of a clinical trial or research study. The declaration of Helsinki is thus not applicable and approval of the ethics committee is not required.

Sections from formalin-fixed, paraffin-embedded tumor were stained with hematoxylin and eosin or alician blue. Pathology revealed a 60-mm reddish gelatinous lobulated antral mass (Fig. 1A), involving submucosa, muscularis propria, and subserosa; the overlying mucosa was ulcerated. Histology showed a plexiform tumor composed of cells with oval nuclei, indistinct cytoplasm and, occasionally, clear halos, in a myxoid, alician-positive matrix, sometimes with tiny collagen bundles, with arborizing capillary vessels (Fig. 1B and C). Tumor cells were positive for α-SMA (Fig. 1D), vimentin (not shown) and, partially, for caldesmon (Fig. 1E), desmin and CD10 (not shown); moreover, focal positivity for AE1/AE3 (Figs. 1F) and pan-Ck KL1 (not shown) was detected. CD117, DOG1, CD34, S100, CAM5.2, CK20, CK7, chromogranin A, synaptophysin, Melan-A, HMB-45, and anaplastic lymphoma kinase (ALK) were all negative (not shown). KIT (exons 9, 11, 13, and 17) and PDGFRA (exons 12, 14, and 18) amplified using the same primers and polymerase chain reaction conditions described elsewhere,[35] were wild type. These findings rule out GIST, the most common mesenchymal tumor of stomach,[36] and carcinoma, because of both morphology and inconsistent immunophenotype; conversely, they are typical of PAMT, with the exception of the focal CK (AE1/AE3 and KL1) expression, exceptional in this tumor type.

3. Discussion

In this study, we report the exceptional occurrence of CK expression in a typical PAMT. PAMT, also known as plexiform fibromyxoma, is a myofibroblastic tumor recently fully characterized.[1,2] Probably the same tumor had been previously signaled several times in the pre-IHC era.[3–7] At the best of our knowledge, 59 PAMTs (including the present case and 2 uncertain ones) have been described in the literature (Table 1).[1,2,8–33] With the caveat due to the limitations in both number of cases and follow up, PAMT appears a benign entity; in fact, neither metastases nor relapses after complete surgical resection have been signaled so far. PAMT is capable of smooth muscle differentiation, as shown by the possible focal expression of caldesmon and desmin.[1,2,12,22,28] As such, PAMT can be expected to occasionally express Ck, since both myofibroblastic and smooth muscle tumors are known to be sometimes able to express these markers.[36] A restrict number of mesenchymal tumors (i.e., synovial sarcoma and epithelial sarcoma) display a true epithelial differentiation, with expression of both low- and high-molecular weight CK isoforms and of other epithelial markers, such as desmoplakins and occludin. Unlike these sarcomas, CK expression found in smooth muscle and myofibroblastic tumors is anomalous and does not reflect genuine epithelial differentiation, involving only a subset of neoplastic cells, mostly with IHC staining limited to a portion only of the cytoplasm, sometimes in a dot-like pattern.[17] This is the case of the herein reported PAMT (Fig. 1F). Coherently with the lack of a true epithelial differentiation, EMA IHC resulted negative, as happened in all PAMTs previously tested for this marker.[10,13,18,19,27,30] with the exception of a focal positivity in a single case reported in the Chinese literature.[20] The presence of a hybrid epithelial-mesenchymal phenotype (so-called “amphicrine pattern”) is a feature of epithelial-to-mesenchymal
and mesenchymal-to-epithelial cell transitions (MET), phenomena which can be found either in organ development or tumors. With regard to gastrointestinal (GI) mesenchymal tumors, MET has been described in GISTs, apparently with a favorable prognostic role. Given the lack of aggressive behavior in the hitherto reported PAMTs, there is no room for a similar biological role of MET in these tumors.

The morphology of the herein reported PAMT (as happens with PAMT as a whole) does not support a diagnosis of carcinoma, although some carcinomas may show myxoid features. In particular, its typical discohesive architecture excludes most carcinomas with the possible exceptions of poorly cohesive gastric carcinoma and gastric metastasis from a lobular breast cancer. However, the former is ruled out by the lack of atypia in the overlying mucosal epithelium and the latter by the clinical context of the reported tumor (lobular breast cancer is very rare in males). Furthermore, both of these neoplasms are excluded by the detected tumoral immunohistochemical profile.

PAMT must be distinguished from other mesenchymal tumors which can be found in the GI tract. GISTs (which can display myxoid or plexiform features) are typically CD117+ and DOG1+, express CD34 in about 2/3 of cases and mostly bear an activating mutation in either KIT or PDGFRA. Inflammatory fibroid polyposis, although often arising in the gastric antrum, rarely grow deeper than submucosa, typically feature CD34+ spindle cells arranged in an onion-skin pattern around blood vessels, are rich in eosinophils and are often PDGRA mutant. Schwannomas, although often displaying areas with a loose texture (so-called “Antoni B areas”) and sometimes featuring a plexiform architecture, often exhibit peripheral lymphoid aggregates and are consistently intensely and diffusely S100+. Inflammatory myofibroblastic tumors feature a relevant inflammatory infiltrate which, together with the loosely arranged myofibroblasts in an edematous myxoid background, simulates granulation tissue; moreover, about half of cases display cytoplasmic positivity for ALK protein. Abdominal desmoid-type fibromatoses feature myofibroblasts arranged in long sweeping bundles, set in a collagenous stroma and, although sometimes showing myxoid change, lack a plexiform architecture and are mostly β-catenin positive at nuclear level. Perhaps the gastric mesenchymal tumor which can be more easily confused with PAMT is myxoid leiomyoma, given its positivity for α-SMA, desmin, and caldesmon; however, it usually arises in the cardias or fundus, and is composed of cells with relatively abundant, intensely eosinophilic cytoplasm, with blunt-ended nuclei and intensely and diffusely desmin+ and caldesmon+. In females, PAMT must be distinguished from metastatic low-grade endometrial stromal sarcoma (ESS); in fact, progesterone receptor positivity has been exceptionally reported in 2 PAMTs, while ESS displays CD10 positivity, can be myxoid and can metastasize to the GI tract; a clinical history negative for gynecological neoplasms and the lack of estrogen and progesterone receptors exclude this entity. Table 2 summarizes the differential diagnosis of PAMT.

Although the PAMT CK expression we report is thus not surprising, given the tissue lineage of this neoplasm, our finding
| Lesion                                    | Morphology                                                                 | Immunophenotype of spindle cells | KIT/ PDGFRA genotype |
|------------------------------------------|-----------------------------------------------------------------------------|----------------------------------|----------------------|
| GIST                                     | Usually centered in the muscularis propria. Spindle and/or epithelioid cells with mildly eosinophilic, often vacuolated, cytoplasm. Variable, mostly low mitotic activity. | + (+~40%) – (5% to 10%+), mostly gastric epithelioid | KIT (75% to 80%) or PDGFRα (5% to 8%) mutant PDGFRα mutant (50-70%) |
| Inflammatory fibroid polyp               | Usually centered in the submucosa, often polypoid. Spindle-ovoid cells in short fascicles, often around blood vessels (usually numerous) in an onion skin pattern; leukocytic infiltrate rich in eosinophils. Low mitotic activity. | + – – – – – (+, often in intestinal cases) | KIT (75% to 80%) or PDGFRα (5% to 8%) mutant PDGFRα mutant (50-70%) |
| GI schwannoma                            | Unencapsulated. Elongated cells; tapering spindled nuclei; ample, ill-delimited eosinophilic cytoplasm; mostly in sheets and interlacing fascicles; palisading, prominent thick-walled/hyalinized blood vessels; lymphoid cells aggregates at the periphery and perivasculatle; foamy macrophages. Low mitotic activity. | – – – – + – | WT |
| Inflammatory myofibroblastic tumor        | Myofibroblasts and inflammatory infiltrate (plasma cells, lymphocytes, and eosinophils) in one of the following patterns: plump or spindled cells loosely arranged in an edematous myxoid background, with abundant blood vessels; compact fascicular spindle cell proliferation with ganglion-like cells with vesicular nuclei and eosinophilic nucleoli in myxoid or collagenized areas, with inflammation diffuse or in small aggregates; scar-like plate-like collagen with low cellularity and relatively sparse inflammatory infiltrate. Low mitotic activity. | + (~90%) + (10% to 70%) – – – – – + 50% to 60%) | WT |
| Abdominal desmoid-type fibromatosis       | Poorly circumscribed, infiltrative proliferation of elongated, slender, spindle-shaped uniform cells, without atypia, in long sweeping bundles in a collagenous stroma with variably prominent often slit-like blood vessels; micromyoinclusions. Variable mitotic activity. | + – (5% +) – – (reported+, depending on technical artifacts) – – – – + (70% to 75%) | WT |
| Smooth muscle tumors                     | Spindled or slightly epithelioid cells with intensely eosinophilic, sometimes vacuolated, cytoplasm, and uniform blunt-ended, cigar-shaped nuclei in intersecting fascicles; leiomyoma mostly paucicellular; may be myxoid change. Significant atypia in leiomyosarcoma. Mitotic activity low in leiomyoma, often high in leiomyosarcoma. | + (~100% leiomyoma, ~70% to 100% leiomyosarcoma) + (~100% leiomyoma, ~50% to 90% leiomyosarcoma) + (~100% leiomyoma, ~50% to 95% leiomyosarcoma) – – – – – (may be + in females) | WT |
| Low-grade endometrial stromal sarcoma,   | Infiltrating masses of uniform, mostly oval small cells resembling endometrial stroma, surrounding small vessels; foci of hyalinization and foamy cells. Variable prominent myxoid change. | + – (sometimes focally +) – – (+ ~5% of cases, focal) – – – – + (40%) | WT |

ALK = anaplastic lymphoma kinase, GANT = gastrointestinal autonomic nerve tumor, GI = gastrointestinal, GIST = gastrointestinal stromal tumor, NA = not assessed, PR = progesterone receptor, SMA = smooth muscle actin, WT = wild type.
nevertheless contributes to enlarge the known immunophenotypic spectrum of this tumor. In fact, at the best of our knowledge, all PAMTs so far immunohistochemically tested for CK expression resulted negative[1,2,3,13,15,16,18,22,24,28,29] with the exception of the report of focal positivity in a single case from the Chinese literature.20,21 But, beyond its descriptive value, the awareness of a possible CK positivity in PAMT is relevant for avoiding possible misinterpretations of PAMT biopsies, especially when dealing with suboptimal amounts of tissue. In fact, under these circumstances, there is the risk of misdiagnosing PAMT as poorly cohesive gastric carcinoma, a neoplasm which can be extensively infiltrative in spite of mucosal lesions endoscopically elusive, potentially leading to dramatic consequences in the management of patients with PAMT.

In conclusion, we demonstrate the possible expression of CK in PAMT, an exceptional finding which, although not surprising considering the tissue lineage of this tumor, can be very relevant in the routine practice of pathologists for avoiding possible misdiagnoses with heavy clinical consequences.

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