Case Report

A Pancreatic Inflammatory Myofibroblastic Tumor with Spontaneous Remission: A Case Report with a Literature Review

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Abstract: The inflammatory myofibroblastic tumor (IMT) is a rare tumor that can develop in any systemic organ. Its features are generally benign, but it often resembles malignancies and is treated surgically. Our patient was an 82-year-old female complaining of abdominal discomfort. Computed tomography demonstrated a 5 cm, ill-enhanced mass at the pancreas head. Upper gastrointestinal endoscopy revealed a duodenal submucosal tumor with apical erosion. Endoscopic ultrasonography (EUS) demonstrated a heterogeneous, low-echoic pancreas mass without clear margins. Fine-needle aspiration biopsy (FNAB) demonstrated spindle myofibroblastic tissues with lymphoplasmacyte and eosinophil infiltration, confirming an IMT diagnosis. Surprisingly, the tumor spontaneously regressed in one month without medication. Histological diagnosis using EUS-FNAB is essential for the rare pancreatic solid tumor like IMT.

Keywords: pancreas; inflammatory myofibroblastic tumor; spontaneous regression; diagnosis

1. Introduction

The inflammatory myofibroblastic tumor (IMT) is a rare tumorous lesion that can develop in any systemic organ. It has a relatively young onset (mainly in newborns to young adults) and consists histologically of proliferative myofibroblastic tissues with heavy infiltration of inflammatory cells, mainly lymphocytes and plasma cells [1]. It often shows benign biological behaviors, but it is sometimes accompanied by somatic mutations in clinically important genes [2-5] that can cause metastasis and/or recurrence [25]. The differential diagnosis from malignancies is therefore difficult, especially in high-aged cases, and most IMTs are surgically resected before finally being diagnosed [2,5–26]. Anti-inflammatory agents, such as corticosteroids and non-steroidal anti-inflammatory drugs (NSAIDs), are effective for some IMTs [27,28], and a small proportion of IMTs regress spontaneously [27–38]. To date, several cases of pancreatic IMTs have been reported [6–26], but histologically proven cases (proven for myofibroblastic component) with spontaneous remission have hardly been reported.

2. Case Presentation

An 82-year-old Japanese female was referred to our hospital after a month of complaints of upper abdominal discomfort. First, she visited the nearest hospital and underwent upper gastrointestinal endoscopy that pointed multiple erosions and an extrinsic compression at the posterior pylorus.
Medication had been initiated using nizatidine, rebamipide and oxetacaine, but it was not effective. She had a history of hypertension, but her family history was unremarkable. Blood tests showed modestly elevated IgG (1950 mg/dL, normal range: 870–1700 mg/dL) and C-reactive protein (0.43 mg/dL, normal range: ≤ 0.3 mg/dL) but normal readings for other factors, including serum tumor markers (carcinoembryonic antigen, carbohydrate antigen, and soluble IL-2 receptor), HbA1c, and IgG4 (66.1 mg/dL, normal: 4.5–117 mg/dL). Enhanced computed tomography (CT) demonstrated an ill-enhanced mass, 5 cm in size but with unclear margins, located at the pancreas head (Figure 1a,b). Upper gastrointestinal endoscopy revealed a submucosal tumor (SMT) with an apical erosion approximately 1.5 cm in size at the duodenal bulbs (Figure 2). Several faintly enlarged lymph nodes were seen around the pancreas head, but no nodules suggestive of metastasis were visible in the liver or the lungs. Endoscopic ultrasonography (EUS) demonstrated a heterogeneous, low-echoic mass at the pancreas head and body, but no adhesion to the common bile duct. EUS elastography revealed a hardness of the pancreas lesion (Figure 3). Forceps biopsy (Radial Jaw™4, Boston Scientific Japan, 2.2 mm, Tokyo, Japan) from the duodenal SMT was not informative, but EUS-guided fine needle aspiration biopsy (FNAB) showed abundant spindle myofibroblast tissues with eosinophilic and lymphoplasmacytic cell infiltration (Figure 4). FNAB was performed with two punctures from the duodenal bulbs, with each puncture performed with 10 strokes using a 22-gauge Franseen-tip needle (Acquire™, Boston Scientific Japan) with 10 mL of negative pressure. No malignant cells were seen. The spindle cells were positive for anti-smooth muscle antibody (ASMA) and desmin but negative for discovered on GIST-1 (DOG-1), c-Kit, CD34, S-100, and ALK. Only six IgG4-positive cells were recognized in high-powered views, and no obliterative phlebitis or storiform fibrosis was detected. These findings led to the diagnosis of IMT.

Figure 1. Enhanced computed tomography. An irregular-margined, low-attenuated mass 5 cm in size was seen at the pancreas head (horizontal view (a), coronal view (b)). One month after the histological diagnosis, the pancreatic mass was markedly shrunken spontaneously (horizontal view (c), coronal view (d)).
Figure 2. Endoscopic view of the duodenal bulbs. A hemispheric submucosal tumor with apical erosion was recognized.

Figure 3. Endoscopic ultrasonography (EUS) views. A heterogeneous low-echoic mass was seen by scanning from the duodenal bulbs (a). Elastography showed a heterogeneously hard mass lesion at the pancreas head (b).

Ten days after FNAB, positron emission tomography showed abnormal $^{18}$F-fluorodeoxyglucose uptake (SUVmax: 6.95); however, the pancreatic lesion seemed to have shrunk to 2.5 cm in size (Figure 5). Magnetic resonance imaging (MRI) demonstrated an obviously minimized tumorous lesion at the pancreas head (Figure 6). The mass lesion was visible as an iso-intensity signal in a T1-weighted image and as a faintly low-intensity signal in a T2-weighted image, while it was ill enhanced in an EOB image and the signal was heterogeneously repressed in a diffusion-weighted image. A subsequent CT, conducted one month after the FNAB, revealed further minimization of the pancreatic mass (Figure 1c,d). The images obtained in the next two months showed that the tumor had almost vanished. The tumor was no longer visible at the sixth month. During the post-diagnosis course, no medication was administered other than regularly taken hypotensive drugs. A written informed consent was obtained from the patient.
Figure 4. Tissues obtained by EUS-guided fine needle aspiration biopsy (EUS-FNAB). Low-power view of hematoxylin-eosin (HE) staining showed mixed components of dense myofibroblastic tissues and aggregated inflammatory cells (×40). (a) Anti-smooth muscle actin was diffusely positive in the myofibroblastic components (×40). (b) High-power views showed a myofibroblastic cell component (c) and an inflammatory cell component (d) without malignant cells (HE, ×100).

Figure 5. 18F-fluorodeoxyglucose positron emission tomography (FDG-PET). A strong uptake of FDG was visible at the pancreas head (SUVmax: 6.95); however, this looked smaller when compared with the initial computed tomography (CT) image. The one unit of under bar is indicating 1cm and total bar length is 25 cm.
Figure 6. Magnetic resonance imaging (MRI) at two weeks after EUS-FNAB and three weeks after the initial CT. A T1-weighted MRI showed an iso-intensity signal (a), a T2-weighted image showed a faint low-intensity signal (b), a gadoxetate sodium-injected MRI demonstrated a slightly weak enhancement (c), and a diffusion-weighted MRI revealed heterogeneously repressed diffusion ability at the pancreatic lesion (d).

3. Discussion

Our English literature survey on PubMed using the keywords “inflammatory myofibroblastic tumor” with “pancreas” or “pancreatic” listed 167 citations. By excluding other inflammatory pancreatic pseudotumors (such as other mass-forming pancreatitis and autoimmune pancreatitis) and histology-unproven cases, 27 cases of histologically confirmed IMTs (positive for histologically proven myofibroblastic tissue) were listed (Table 1). The classical term “inflammatory pseudotumor (IPT)” often includes other etiologies, such as IgG4-related, autoimmune-related, and infection-causing inflammatory lesions [39]; therefore, we only listed the histologically proven pancreatic IMTs and not the IPTs. Including the current case, the mean age at diagnosis of all cases was 40.0 years, and a subtle predominance of male gender was evident (17 males and 11 females). The pancreatic mass lesions were mostly located in the pancreas head (20 in the head, two in the body, four in the tail, and two in the body and tail) and had a mean size of 4.7 cm. Abdominal pain or discomfort was the most frequent symptom (56%, 15/27), with jaundice the second most frequent (44%), followed by anorexia or weight loss (26%), and nausea or vomiting (15%). One case had stable disease, one case had a recurrence in the lung, and one case died of sepsis, but the other 22 (88%) cases did not show recurrence after resection or remission (Table 1).
### Table 1. Reported cases of inflammatory myofibroblastic tumor (IMT) of the pancreas (English literature).

| No | Ref no | Author | Year | Age (years old) | Gender | Location | Tumor Size (cm) | Symptoms | Pathological Examination (pathology) | Preoperative Diagnosis | Treatment | Course after Surgery or Remission | Follow-up Period | Status |
|----|--------|--------|------|-----------------|--------|----------|---------------|----------|-------------------------------------|----------------------|-----------|---------------------------------|------------------|--------|
| 1  | 26     | Kroft  | 1995| 42              | F      | Pb       | 7             | abd. pain, weight loss, fatigue | FNAB (benign pancreatic tissue) | ND | resection | ND | 6 months | NER |
| 2  | 6      | Shankar| 1998| 8               | F      | Pb       | 10.7          | abd. pain | ND | sarcoma | resection | 2 years | NER |
| 3  | 7      | Walsh  | 1998| 35              | M      | Pb       | 5             | jaundice, abd. pain, anorexia, weight loss | ND | pancreatic cancer | resection | 6 years | NER |
| 4  | 8      | McClain| 2000| 11              | F      | Pb       | 3.4           | jaundice, abd pain, weight loss | ND | malignancy | resection | ND | NER |
| 5  | 9      | Wreesman| 2001| 62              | M      | Pb       | 1.7           | jaundice | ND | pancreatic cancer | resection | 5 years | NER |
| 6  | 10     | Yamamoto| 2002| 55              | M      | Pb       | 1.5           | none (incidental finding) | ND | pancreatic cancer | resection | 12 years | NER |
| 7  | 11     | Esposito| 2003| 69              | M      | Pb       | 3.8           | none (incidental finding) | ND | (no malignancy) | resection | 7 months | died of sepsis |
| 8  | 12     | Pungpapong| 2004| 70              | M      | Pt       | 2             | none (incidental finding) | ND | (no malignancy) | resection | 10 months | NER |
| 9  | 13     | Dulundu| 2007| 65              | M      | Pb       | 2             | none (incidental finding) | ND | ND | resection | 3 years | NER |
| 10 | 14     | Sim    | 2008| 56              | M      | Pt       | 7             | melena | ND | pancreatic cancer | resection | 1.5 years | NER |
| 11 | 15     | Dagash| 2009| 13              | F      | Pb       | 3             | jaundice, vomiting, weight loss | open bp ¥ (IPT) | ND | resection | ND | 6 years | NER |
| 12 | 16     | Hassan| 2010| 19              | M      | Pb       | 2.2           | jaundice, abd. pain, anorexia | percutaneous bp (IPT) | IPT | prednisolone, cefuroxime | 6 years | NER |
| 13 | 17     | Schutte| 2010| 44              | F      | Pb       | 8.2           | abd. pain | ND | splenic rupture | resection | 6 years | NER |
| 14 | 18     | Lacoste| 2012| 56              | M      | Pb       | 6             | abd. pain, nausea, weight loss | bp (not diagnostic) | possible malignancy | resection | 1 year | SD |
| 15 | 19     | Panda  | 2015| 32              | F      | Pb       | 4.8           | jaundice, abd. pain | US-FNAB (not diagnostic) | possible malignancy | resection | 3.5 years | NER |
| 16 | 20     | Zarchi | 2015| 13              | F      | Pb       | 2.5           | jaundice, abd. pain, vomiting, anorexia | US-FNAB (mesenchymal neoplasm) | pancreatic cancer | resection | 2.5 years | NER |
| 17 | 21     | Battal | 2016| 46              | M      | Pb       | 8             | abd. pain | ND | ND | resection | ND | NER |
| 18 | 22     | Ding   | 2016| 69              | M      | Pb       | 4             | vomiting, anorexia | endoscopic bp. (fibrinous lesion with inflammatory cells) | malignancy | resection | 3 years | NER |
| 19 | 23     | Liu    | 2017| 15              | M      | Pt       | 5             | abd. pain | US-FNAB (compatible with IMT) | tumor invading the transverse colon | Resection # | 3 years | NER |
| 20 | 24     | Berhe  | 2019| 1               | F      | Pb       | ND            | abd. pain | ND | IMT or chronic pancreatitis | resection | ND | NER |
| 21 | Current case | 2019 | 82         | F      | Pb       | 5             | abd. discomfort | IMT | IMT | None * | 9 months | NER |

Average M:F = 39.7:17.11, Ph:Ph:Pb:Pt:Pbt: = 20:2:4:2:4.7

| M: male, F: female, ND: not described, NER: no evidence of recurrence, SD: stable disease, Ph: pancreas head, Pb: pancreas body, Pt: pancreas tail, Pbt: pancreas body to tail, IMT: inflammatory myofibroblastic tumor, IPT: inflammatory pseudotumor, abd pain: abdominal pain, ¥ bp: biopsy, US-FNAB €: abdominal ultrasound-guided fine needle aspiration biopsy, # tumor resection and segmental colorectomy, € incidentally detected by the health check images, * Current case showed spontaneous regression without medication. |
These data for pancreatic, IMTs differ greatly from the data of 730 IMT cases summarized by Nonaka et al. [1] who reported a younger onset (mean 29.6 years, range: 0–87 years), a nearly equal gender ratio, favored organs (commonest in lung, followed by the urinary bladder, mesentery, omentum, retroperitoneum, pelvis, gastrointestinal tract, and liver), larger size (mean 5.9 cm, range 0.4–36 cm), varied symptoms (strongly related to the location, but 19% were accompanied by systemic symptoms including fever, malaise, weight loss, and anemia), and different outcomes (local recurrence: 22%; metastasis: 3%; death from disease: 2%; and no recurrence: 67%). This trend became more apparent when comparing 59 cases of systemic IMTs with histological atypia [5], as these had younger onset (mean age: 13.2 years old, ≥20 years old: 29%), an even gender ratio (29 male and 30 female), large tumor size (mean: 7.8 cm), similar location (abdomen and pelvis: 64%, lung: 22%, head and neck: 8%, and extremity: 5%), and poorer outcomes (recurrence: 56%; metastasis: 10%; death from disease: 10%; and no recurrence: 42% in an average of 3.6 years of follow up). Thus, a wide range of variation exists in the nature of IMTs, and the adult pancreatic IMTs may have a relatively benign nature.

The biological marker of IMTs, including histological atypia, ganglion-like cells, TP53 expression, and aneuploidy pattern, have been correlated with more aggressive clinical behavior [40]. Coffin et al. also suggested that ALK (anaplastic lymphoma kinase) expression is another prognostic indicator of IMTs. The ALK (2p23) gene is activated by gene rearrangement in 50–70% of IMTs and ALK gene rearrangement is correlated with ALK expression, as determined by immunochemistry [41]. The absence of ALK expression in IMT was associated with a higher age of the patients [5]. All six of the observed metastases developed in 59 IMTs that were negative for ALK expression, and they developed before 20 years of age (mean age: 13.2 years), indicating a metastatic potential for ALK-negative IMTs in the younger subset [5]. The current case of a pancreatic IMT was in a patient of high age (82 years) and showed no histological atypia, ganglion-like cells, or ALK expression. Therefore, ALK expression as a clinical indicator in IMTs in older patients needs further evaluation.

IMTs show spontaneous regression in a minor fraction of patients, although the actual incidence is unclear due to surgical interventions and asymptomatic/undetected cases. To date, 13 cases of spontaneously regressed IMTs have been reported in various organs, but not in the pancreas (Table 2) [27–38]. As mentioned above, reported cases of pancreatic pseudotumors or those without proven myofibroblastic tissues were not listed [15,23]. Of the 13 cases, corticosteroids and/or NSAIDs were used in six cases and were effective in five cases. This phenomenon leads us to question the neoplastic nature of this tumor. IMTs often expand in size to invade multiple organs; therefore, when an accurate diagnosis can be made, conservative treatments should be recommended for older patients.
Table 2. Reported cases of inflammatory fibroblastic tumor (IMT) with spontaneous and/or drug-used remission (English literature).

| No. | Ref. no. | Author         | Year | Age (years old) | Gender | Tumor Size (cm) | Location                | Symptoms                        | Histological Examination | Treatment                                                                 | Course after Remission |
|-----|----------|----------------|------|-----------------|--------|-----------------|--------------------------|--------------------------------|------------------------------|----------------------------------------------------------------------------|------------------------|
| 1   | 27       | Przkora        | 2004 | 63              | F      | ND              | retroperitoneum and        | none                           | ND                           | prednisolone: 150 mg/day and dexamethasone 50 mg × 2/day for 1 week | 14 months, NER       |
| 2   | 22       | Mab             | 2008 | 28              | M      | ND              | skull base                | hearing loss, headache,       | open bp                      | none                                                                      | 3 years, NER           |
| 3   | 29       | Galindo        | 2008 | 28              | M      | ND              | mediastinum               | abd. discomfort, anorexia     | US-guided bp                  | right lower lobectomy (none for left lung lesion)                       | 4 months, NER          |
| 4   | 30       | Mattei         | 2008 | 13              | M      | ND              | duodenum                 | abd. pain, vomiting, weight  | needle bp, lung lobectomy    | none                                                                      | 1 year, NER            |
| 5   | 31       | Sugiyama        | 2008 | 72              | M      | ND              | mediastinum               | hearing loss, headache,       | open bp                      | none                                                                      | 1 year, NER            |
| 6   | 32       | Bilaceroglu     | 2009 | 21              | F      | 6, 2            | bilateral lung            | abd. pain, vomiting, weight  | none (anemia)                 | antibiotics                  | 6 years, NER             |
| 7   | 33       | Fragoso         | 2011 | 14              | M      | ND              | diffuse involvement of     | liver                         | FNAB                         | prednisone 20 mg/day and celecoxib 200 mg/day for 2 weeks                | 3 months, NER          |
| 8   | 34       | Shatzel         | 2012 | 28              | F      | 5.5             | mesentery                | abd. pain                     | incomplete mass resection     | ND, NER                     | 3 months, shrunk to 4.2 cm |
| 9   | 35       | Calaway         | 2014 | 71              | F      | 5               | kidney                   | abd. pain, vomiting, fever   | percutaneous FNAB (IMT)        | none                        | 5 years, NER             |
| 10  | 36       | Zhao            | 2014 | 49              | M      | 15              | retroperitoneum           | abd. pain, vomiting, fever   | laparoscopic incisional bp    | none                        | 3 months, NER             |
| 11  | 37       | Markovic        | 2016 | middle age      | F      | 3.9             | gastric wall             | abd. distension, weight loss  | laparotomy, lymphadenectomy   | none                        | 1 year, NER              |
| 12  | 38       | Yoshimura       | 2016 | 78              | F      | ND              | cauda equina             | pain and numbness in buttock | laminectomy; intraoperative  | none                        | 3 years, NER              |
| 13  | 39       | Habib           | 2017 | 7               | M      | 1.7             | orbit                    | decreased visual acuity,      | orbital bp                    | corticosteroid (failure)     | 12 years, shrunk to 0.8 cm |
| 14  | 40       | Current case    | 2019 | 82              | F      | 5               | pancreas                 | abd. discomfort               | EUS-FNAB                     | none                        | 9 months, NER             |

Average 43.4 (8:6) 5.6

M: male, F: female, ND: not described, NER: no evidence of recurrence, SD: stable disease, EUS: endoscopic ultrasonography, FNAB: fine needle aspiration biopsy, bp: biopsy, IMT: inflammatory myofibroblastic tumor.
An accurate diagnosis of pancreatic IMTs cannot be made by image examinations alone, as most cases mimic malignancies (Table 1) and no IMT serum markers are commercially available. EUS-FNA demonstrates a fairly high diagnostic ability (nearly 95% sensitivity and specificity) for solid malignant pancreatic lesions [42,43]. The use of thick core biopsy needles [44] and high-negative-pressure aspiration methods [45] has increased the acquisition rate for obtaining core tissue samples. This, in turn, has enabled the determination of the probable nature of the whole pancreatic mass and even the classification of intermediate inflammatory and neoplastic conditions, such as IMTs. In the present case, we performed a conventional EUS-FNA but used a 22-gauge Franseen needle, and our pathologist was able to diagnose IMT (Figure 4). Other similar conditions, such as other inflammatory pseudotumors (IgG4-related [39,46], autoimmune-related [47], and infection-related [39] masses) and spindle cell tumors (malignant fibrous histiocytoma [47], sarcomatoid anaplastic large cell lymphoma, spindle cell carcinoma, inflammatory leiomyosarcoma, and pleomorphic liposarcoma) [5], must be carefully ruled out by multiple immunohistochemical tests.

In conclusion, we have reported a rare case of pancreatic IMT demonstrating spontaneous remission. Our aim was to emphasize the importance of accurate diagnosis that includes histology obtained by EUS-FNA. Further accumulation of cases is needed to clarify the biological behavior of pancreatic IMTs.

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Abbreviations

| Abbreviation | Description |
|--------------|-------------|
| IMT | inflammatory myofibroblastic tumor |
| NSAIDS | non-steroidal anti-inflammatory drugs |
| CT | computed tomography |
| SMT | submucosal tumor |
| EUS | endoscopic ultrasonography |
| FNAB | fine needle aspiration biopsy |
| ASMA | anti-smooth muscle antibody |
| MRI | magnetic resonance imaging |
| IPT | inflammatory pseudotumor |
| ALK | anaplastic lymphoma kinase |

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