Inflammatory myofibroblastic tumor of the pancreatic neck: A case report and review of literature

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Abstract

BACKGROUND
Pancreatic inflammatory myofibroblastic tumor (IMT) is a relatively rare disease that is often confused with pancreatic cancer or pancreatic neuroendocrine tumors. The histological features of IMTs show that tissue from this type of tumor contains an intermingling of fibroblast and myofibroblast proliferation, accompanied by a varying degree of inflammatory cell infiltration.

CASE SUMMARY
The management of an IMT occurring at the neck of the pancreas is presented in
this paper. A 66-year-old female patient was diagnosed with a pancreatic neck mass after a series of tests. The patient underwent enucleation of the pancreatic neck tumor after a pathological diagnosis of IMT. Previous research on the clinical features, pathological diagnosis and treatment of pancreatic IMTs was reviewed. Compared with previous reports, this is a unique case of enucleation of a pancreatic IMT.

CONCLUSION

The enucleation of pancreatic IMTs may be a safe and efficient surgical method for managing such tumors with a better prognosis. Further cases are required to explore surgical measures for pancreatic IMTs.

Key Words: Inflammatory myofibroblastic tumor; Pancreatic neck; Enucleation; Case report

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INTRODUCTION

An inflammatory myofibroblastic tumor (IMT) is a rare mesenchymal tumor of unknown pathogenesis and aggressive malignant potential with a global incidence of less than 1% [1,2]. IMTs most commonly occur in the lungs of children and young adults, followed by the head and neck [3], liver [4], pancreas [5], genitourinary tract [6] and thyroid [7]. The clinical presentation of pancreatic IMTs varies depending on their anatomic location, and the final diagnosis of most lesions requires a pathological examination. The pancreatic head is the most common site for pancreatic IMTs and may be the first choice for surgical resection. Of 29 cases of pancreatic IMT reported in the English literature, none have been treated by enucleation of the tumor. Herein, an unusual pancreatic neck IMT occurring in a 66-year-old female patient is presented, and this may be the first case of enucleation of a pancreatic IMT. Pancreatic IMTs have a relatively low incidence and unspecific manifestations. The clinical and histological features of pancreatic IMTs, as well as their diagnosis and treatment, are discussed in this paper.

CASE PRESENTATION

Chief complaints

A 66-year-old female patient was admitted to Shulan (Hangzhou) Hospital on January 13, 2020 for a pancreatic mass.

History of present illness

Abdominal ultrasonography of the patient showed hyperechoic foci in the neck of the
pancreas after a follow-up examination in the local hospital 4 d prior, and then the patient was transferred to our department for further treatment.

**History of past illness**
The patient had a history of right pulmonary wedge resection for adenocarcinoma in 2014 and right hemicolectomy for colon cancer in 2018.

**Physical examination**
The physical examination was unremarkable.

**Laboratory examinations**
Laboratory examinations, including complete blood count, C-reactive protein and tumor markers, were all within the normal range.

**Imaging examinations**
However, the ultrasound scan revealed a 2.5 cm × 1.5 cm mass in the neck of the pancreas. Dynamic contrast-enhanced magnetic resonance imaging scan showed an abnormal soft tissue heterogeneous mass in the neck of the pancreas, which appeared hyperintense on the T1-weighted image and mildly hyperintense on the T2-weighted image. A centripetal enhancement pattern was observed during the delayed phase of contrast imaging (Figure 1A-E). Whole-body 18F-fluorodeoxyglucose (FDG) positron emission tomography/computed tomography (CT) examination revealed a 2.3 cm × 1.4 cm, mild-to-moderate FDG uptake nodule in front of the pancreatic neck (SUVmax 3.87) with normal scans of the head, neck, chest and colon (Figure 1F). The imaging findings were highly suggestive of pancreatic IMT. However, the possibility of a metastatic tumor could not be ruled out due to the history of lung and colon cancer.

**Histopathological examination**
A detailed postoperative histopathological examination revealed that the carcinoma cells stained positively for desmin, vimentin, CD34, CD31, BCL2 and β-catenin and negatively for S-100, Pan-CK (AE1/AE3), caldesmon, DOG1, CD117, smooth muscle actin and P53.

**FINAL DIAGNOSIS**
A diagnosis of pancreatic neck IMT was determined on the basis of the histopathological results (Figure 2).

**TREATMENT**
The patient with pancreatic IMT underwent enucleation of the pancreatic mass after multidisciplinary team discussion. During the laparotomy, a hard protruding mass with a size of 2.3 cm × 1.5 cm was observed on the pancreatic neck and subsequently enucleated. The entire mass was fleshy with a grayish-white cut surface and was confirmed with the intraoperative frozen section to be an IMT.

**OUTCOME AND FOLLOW-UP**
The postoperative recovery was uneventful, and the patient was discharged on postoperative day 11 (Figure 3). No adjuvant treatment was administered, and no obvious signs of metastasis or recurrence in the next 10 mo of follow-up were observed.

**DISCUSSION**
IMT, first reported in the lungs[8,9], is a special type of disease that is often termed differently in primary research, including designations such as plasma cell granuloma, plasma cell pseudotumor, inflammatory pseudotumor, inflammatory fibroxanthoma and histiocytoma[10]. IMTs can occur almost anywhere in the body, including the
lungs, liver, bladder, mesentery and neck[11-13]. However, an IMT arising from the pancreas is extremely rare. A complete search of the literature from 1900 to 2020 using the PubMed database with the search terms “inflammatory myofibroblastic tumor,” “IMT,” “pancreas” and “pancreatic” was performed, and only 29 reported cases were discovered. A brief literature review of reported cases with pancreatic IMT was conducted to better understand pancreatic IMT, as summarized in Table 1[5,10,14-34]. Of these patients, 20 were male (20/29, 69%), and 9 were female (9/29, 31%), with an obvious male predilection. The tumor diameter for all reported cases ranged from 1.5 to 15.0 cm. Most tumors occurred in the pancreatic head (21/29 patients), followed by the pancreatic tail (4/29 patients) and pancreatic body (3/29 patients), suggesting that
| Cases | Sex | Age in yr | Location | Diameter in cm | Symptoms | Treatment | Follow-up | Ref. |
|-------|-----|-----------|----------|----------------|----------|-----------|-----------|------|
| 1     | M   | 70        | PT       | 3.8            | Asymptomatic | DP + splenectomy | Disease-free at 10 mo | Pungpaypon et al [29], 2004 |
| 2     | M   | 62        | PH       | 3              | Jaundice     | PD        | Disease-free at 6 yr | Wereesmann et al [14], 2001 |
| 3     | M   | 56        | PH       | no             | Jaundice     | PD        | Disease-free at 5 yr | Wereesmann et al [14], 2001 |
| 4     | M   | 50        | PH       | 5              | Jaundice, abdominal pain | PD    | Disease-free at 4 yr | Wereesmann et al [14], 2001 |
| 5     | F   | 57        | PH       | Not available  | Jaundice     | PD        | Disease-free at 3 yr | Wereesmann et al [14], 2001 |
| 6     | M   | 45        | PH       | Not available  | Jaundice     | PD        | Disease-free at 10 yr | Wereesmann et al [14], 2001 |
| 7     | F   | 32        | PH       | 3              | Abdominal pain | PD        | Disease-free at 12 yr | Wereesmann et al [14], 2001 |
| 8     | F   | 42        | PB       | 7              | Abdominal pain, weight loss | PD    | Disease-free at 6 mo | Kroft et al [15], 1998 |
| 9     | F   | 8         | PBT      | 10.7           | Abdominal mass | PD       | Disease-free at 2 yr | Shankar et al [16], 1998 |
| 10    | M   | 35        | PH       | 5 × 4 × 3      | Abdominal pain, weight loss | PD    | Lung metastasis at 6 yr | Walsh et al [17], 1998 |
| 11    | M   | 55        | PH       | 1.5            | Asymptomatic  | PD        | Disease-free at 28 mo | Yamamoto et al [18], 2002 |
| 12    | M   | 69        | PBT      | Not available  | Abdominal pain | DP + splenectomy + colon splenic flexure | Died after 7 mo of hospitalization due to sepsis | Esposito et al [19], 2004 |
| 13    | M   | 65        | PB       | 2              | Asymptomatic  | DP + splenectomy | Disease-free at 3 yr | Dulundra et al [19], 2007 |
| 14    | M   | 56        | PT       | 5 × 7          | Melena       | DP + splenectomy | Disease-free at 18 mo | Sim et al [30], 2008 |
| 15    | F   | 13        | PH       | 3              | Vomiting, weight loss | PD       | Disease-free at 7 yr | Dagash et al [20], 2009 |
| 16    | M   | 10        | PH       | 2.2            | Abdominal pain, anepithymia | Prednisolone, cefuroxime | Disease-free at 6 yr | Dagash et al [20], 2009 |
| 17    | M   | 19        | PT       | 8.2 × 6.5 × 6.0| Abdominal pain | DP + splenectomy | Disease-free at 6 yr | Hassan et al [22], 2010 |
| 18    | M   | 44        | PH       | 6 × 4          | Abdominal pain, vomiting | PD       | Disease-free at 1 yr | Chütte et al [23], 2010 |
| 19    | M   | 65        | PH       | Not available  | Abdominal pain | PD       | Not available | Lacoste et al [25], 2012 |
| 20    | M   | 0.5       | PH       | 4              | Jaundice     | PD        | Disease-free at 3.5 yr | Tomazic et al [31], 2015 |
| 21    | F   | 32        | PH       | 4.8 × 3.2      | Abdominal pain | PD        | Disease-free at 2.5 yr | Panda et al [26], 2015 |
| 22    | M   | 46        | PH       | 8 × 6 × 5      | Jaundice     | PD        | Not available | Battal et al [27], 2016 |
| 23    | M   | 69        | PH       | 4 × 3          | Vomiting, anepithymia | PD       | Disease-free at 3 yr | Ding et al [21], 2016 |
| 24    | M   | 15        | PH       | 5 × 5 × 4.3    | Abdominal pain, fever | PD       | Not available | Liu et al [24], 2017 |
| 25    | M   | 1         | PH       | 4 × 3          | Asymptomatic  | PD       | Not available | Berhe et al [34], 2019 |
| 26    | F   | 82        | PH       | 5              | Abdominal pain | None     | Disease-free at 9 months | Matsubayashi et al [28], 2019 |
Clinical manifestations
Pancreatic IMT can occur at all ages but shows a preference for children and young adults[35]. All reported cases range from 6 mo to 82 years (mean age: 42 years). As described previously, the clinical presentation of pancreatic IMT varies depending on its anatomic location and can range from asymptomatic to hemorrhagic shock due to rupture of the spleen[19,22]. Nonetheless, almost all pancreatic IMTs have similar nonspecific symptoms, such as abdominal discomfort, abdominal distension, abdominal pain, general fatigue and weight loss. Obstructive jaundice may be noted in typical patients with a pancreatic head IMT. The tumor can also obstruct the pancreatic duct and induce chronic pancreatitis with abdominal discomfort, diarrhea and indigestion[23]. An IMT arising from the pancreatic tail can also obstruct blood vessels of the spleen, resulting in rupture of the spleen with severe abdominal pain and hemorrhagic shock[22]. However, the IMT of our patient arose from the neck of the pancreatic IMT was more common in the pancreatic head.
Clinical evaluation
The preoperative laboratory findings were nonspecific for the diagnosis of pancreatic IMT. Only a few patients with a solitary mass occurring in the head of the pancreas may have elevated total serum bilirubin, amylase and carbohydrate antigen 19-9 due to obstruction of the bile duct or pancreatic duct[26]. Moreover, the radiological features are often deceptive. Ultrasound, CT and magnetic resonance imaging examinations showed mass lesions mimicking pancreatic cancer or pancreatic neuroendocrine tumors. Similar to that of other malignant tumors, whole-body $^{18}$F-FDG positron emission tomography/CT also showed an elevated SUV$_{max}$[36], which can distinguish IMTs from non-neoplastic lesions, such as pancreatic pseudocysts and swollen lymph nodes. In addition, whole-body $^{18}$F-FDG positron emission tomography/CT is the best tool to detect tumor recurrence or distant metastasis. Even standard intraoperative frozen pathology may not provide definitive information to distinguish pancreatic IMTs from pancreatic inflammatory pseudotumors.

Pathology/pathophysiology
The definitive diagnosis of IMTs relies on histological evaluations and immunohistochemical tests[37]. The histological features of IMTs are spindle-shaped cells accompanied by varying degrees of inflammatory cells[38,39]. Coffin et al[37] suggested that clonal cytogenetic abnormalities involving the anaplastic lymphoma kinase gene on the short arm of chromosome 2 at 2p23 occur in approximately 50% of IMTs[37]. This can be a useful test for a definitive clinicopathologic diagnosis. In addition, most extrapulmonary IMTs display immunohistochemical reactivity for spinal muscular atrophy, desmin, the tissue cell marker CD68 and the vascular marker CD34[40].

Treatment
To date, no standard consensus regarding the treatment of pancreatic IMT has been reached. However, surgical resection of the lesion is recommended as the primary therapeutic option for pancreatic IMT. The surgical approach is related to the location of the lesion on the pancreas. For pancreatic head IMTs, pancreaticoduodenectomy is recommended, while distal pancreatectomy is recommended for pancreatic body or tail IMTs. Pancreatic IMTs often invade surrounding organs such as the colon, duodenum and stomach. However, these theories are not widely accepted for such low-grade malignant lesions. Whether radical surgery is necessary requires a large number of further clinical studies.

Radiation therapy, chemotherapy and high-dose steroid therapy have also been used in patients with incomplete resection, impossible resection or malignant disease status postsurgical resection[20,28,41]. Spontaneous regression of pancreatic IMTs has been reported infrequently[28]. Given that our patient was an elderly and infirm female with pancreatic neck IMT only, multidisciplinary team discussion suggested that enucleation would be a more beneficial therapeutic option. No adjuvant treatment
was administered following the enucleation of the pancreatic IMT. The patient remained symptom-free and healthy without tumor recurrence or metastasis 10 mo after surgery. Although only one patient with IMT has been reported to have undergone enucleation, such operative procedures could be considered in the future. More cases are required to explore the surgical treatment of pancreatic IMTs.

**Prognosis**

Pancreatic IMT is regarded as a low-grade malignancy with a generally favorable prognosis. However, a close and long-term follow-up after surgery must be carried out due to its potential for malignancy, distant metastasis and recurrence.

**CONCLUSION**

This paper reports a rare case of IMT of the pancreatic neck managed with enucleation treatment to confirm whether radical surgery could be avoided. This is the first reported case in which enucleation usage resulted in a favorable prognosis of pancreatic IMT. Surgical resection may be the preferred treatment and may provide a better prognosis. However, using enucleation as a surgical measure for treating patients with IMT may also yield a good prognosis.

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