World Journal of Clinical Cases

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World J Clin Cases 2021 August 6; 9(22): 6418-6427

DOI: 10.12998/wjcc.v9.i22.6418

ISSN 2307-8960 (online)

CASE REPORT

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

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Author contributions: Chen ZT and Lin SZ were the patient's surgeons, reviewed the literature and contributed to manuscript drafting; Lin YX reviewed the literature and contributed to manuscript drafting; Li MX performed the interpretation and contributed to manuscript drafting; Zhang T analyzed and interpreted the pathologic findings; Wan DL and Lin SZ performed the disease consultation, reviewed the literature and drafted the manuscript; all authors issued final approval for the version to be submitted.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and

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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

Core Tip: Pancreatic inflammatory myofibroblastic tumor (IMT) is a relatively rare disease that is often confused with pancreatic cancer or pancreatic neuroendocrine tumors. We present herein a 66-year-old female patient who was diagnosed with a pancreatic neck mass after a series of tests. The patient underwent enucleation of a pancreatic neck tumor after pathological diagnosis of IMT. Compared with previous reports, this is a unique case of enucleation of a pancreatic IMT. We conclude that the enucleation of pancreatic IMTs may be a safe and efficient surgical method for managing such tumors with a better prognosis.

Citation: Chen ZT, Lin YX, Li MX, Zhang T, Wan DL, Lin SZ. Inflammatory myofibroblastic tumor of the pancreatic neck: A case report and review of literature. *World J Clin Cases* 2021; 9(22): 6418-6427
URL: https://www.wjgnet.com/2307-8960/full/v9/i22/6418.htm
DOI: https://dx.doi.org/10.12998/wjcc.v9.i22.6418

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

revised according to the CARE Checklist (2016).

Manuscript source: Unsolicited manuscript

Specialty type: Medicine, research and experimental

Country/Territory of origin: China

Peer-review report's scientific quality classification
Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C, C
Grade D (Fair): 0
Grade E (Poor): 0

Received: February 18, 2021
Peer-review started: February 18, 2021
First decision: March 11, 2021
Revised: March 23, 2021
Accepted: June 1, 2021
Article in press: June 1, 2021
Published online: August 6, 2021

P-Reviewer: Cianci P, Tomažič A
S-Editor: Fan JR
L-Editor: Filipodia
P-Editor: Xing YX

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 ^{18}F-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 (SUV_{max} 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

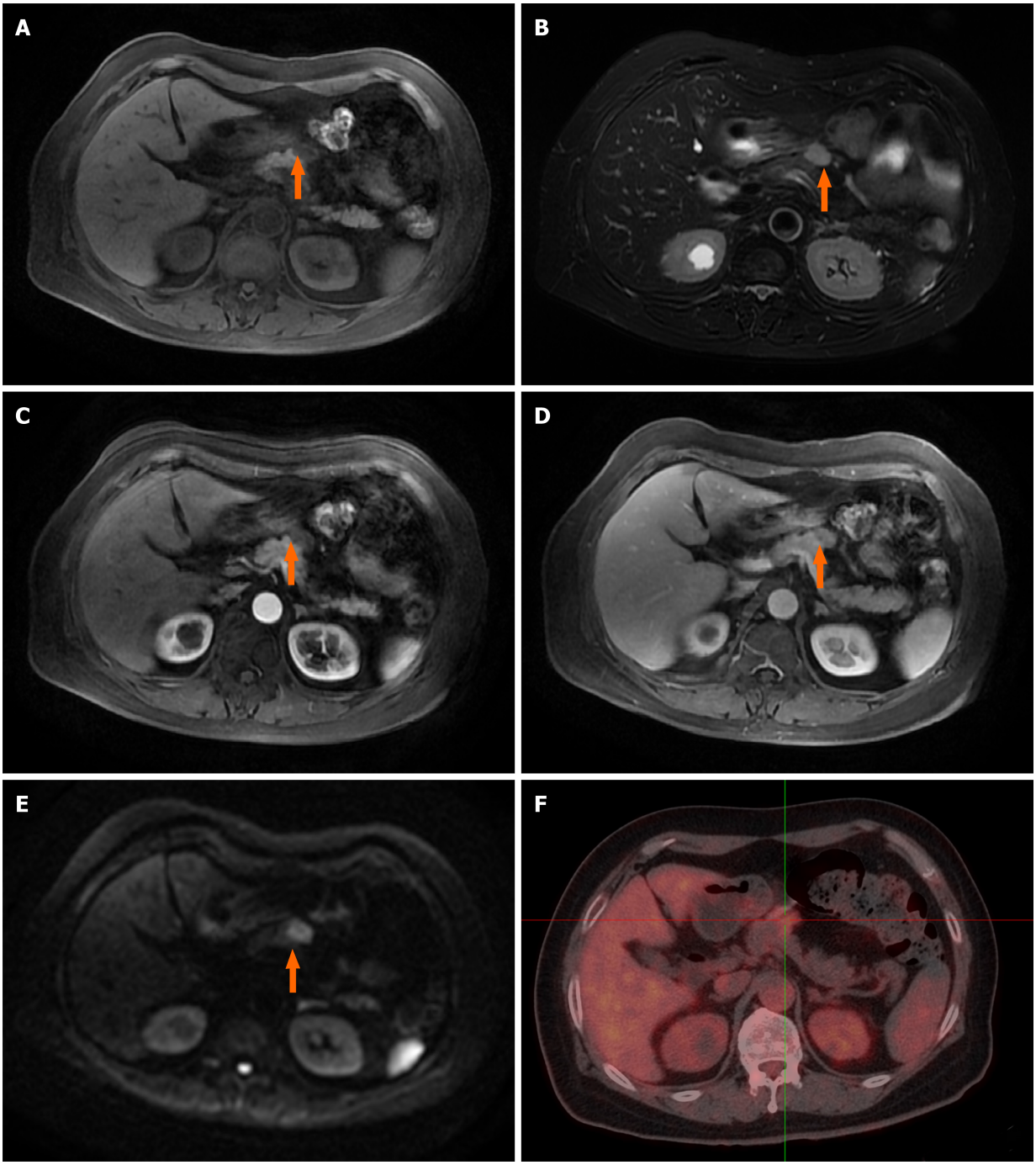

Figure 1 Dynamic contrast-enhanced magnetic resonance imaging and ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography reveal one irregular lesion in the pancreatic neck. A: The lesion of the pancreatic neck (orange arrow) was hyperintense on T1-weighted imaging; B: The irregular lesion (orange arrow) was slightly hyperintense on T2-weighted imaging; C: Arterial phase imaging revealed slight enhancement of the pancreatic lesion (orange arrow); D: The lesion (orange arrow) showed persistent enhancement in venous phase imaging; E: The pancreatic mass (orange arrow) had significant hyperintensity in diffusion-weighted imaging; F: ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography revealed a hypermetabolic pancreatic neck nodule measuring 2.3 cm × 1.4 cm (SUV_{max} = 3.87).

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

Table 1 Reported cases of pancreatic inflammatory myofibroblastic tumor in the English literature

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	Pungpapong *et al* [29], 2004
2	M	62	PH	3	Jaundice	PD	Disease-free at 6 yr	Wreesmann *et al* [14], 2001
3	M	56	PH	no	Jaundice	PD	Disease-free at 5 yr	Wreesmann *et al* [14], 2001
4	M	50	PH	5	Jaundice, abdominal pain	PD	Disease-free at 4 yr	Wreesmann *et al* [14], 2001
5	F	57	PH	Not available	Jaundice	PD	Disease-free at 3 yr	Wreesmann *et al* [14], 2001
6	M	45	PH	Not available	Jaundice	PD	Disease-free at 10 yr	Wreesmann *et al* [14], 2001
7	F	32	PH	3	Abdominal pain	PD	Disease-free at 12 yr	Wreesmann *et al* [14], 2001
8	F	42	PB	7	Abdominal pain, weight loss	DP	Disease-free at 6 mo	Kroft *et al*[15], 1998
9	F	8	PBT	10.7	Abdominal mass	DP	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* [10], 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*[18], 2004
13	M	65	PB	2	Asymptomatic	DP + splenectomy	Disease-free at 3 yr	Dulundu *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	Schütte *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 mouths	Matsubayashi *et al* [28], 2019

27	M	61	PT	15 × 13 × 7	Abdominal pain	DP + left surrenalectomy + left hemicolectomy + splenectomy	Disease-free at 8 mo	İflazoğlu *et al*[5], 2020
28	F	11	PH	3.4	Abdominal pain, weight loss	PD	Not available	Mcclain *et al*[32], 2000
29	F	13	PH	2.5	Abdominal pain	PD	Disease-free at 4 yr	Zanchi *et al*[33], 2015
30	F	66	PN	2.3 × 1.5	Asymptomatic	Enucleation	Disease-free at 9 mo	Current

DP: Distal pancreatectomy; F: Female; M: Male; PB: Pancreatic body; PBT: Pancreatic body and tail; PD: Pancreaticoduodenectomy; PH: Pancreatic head; PN: Pancreatic neck; PT: Pancreatic tail.

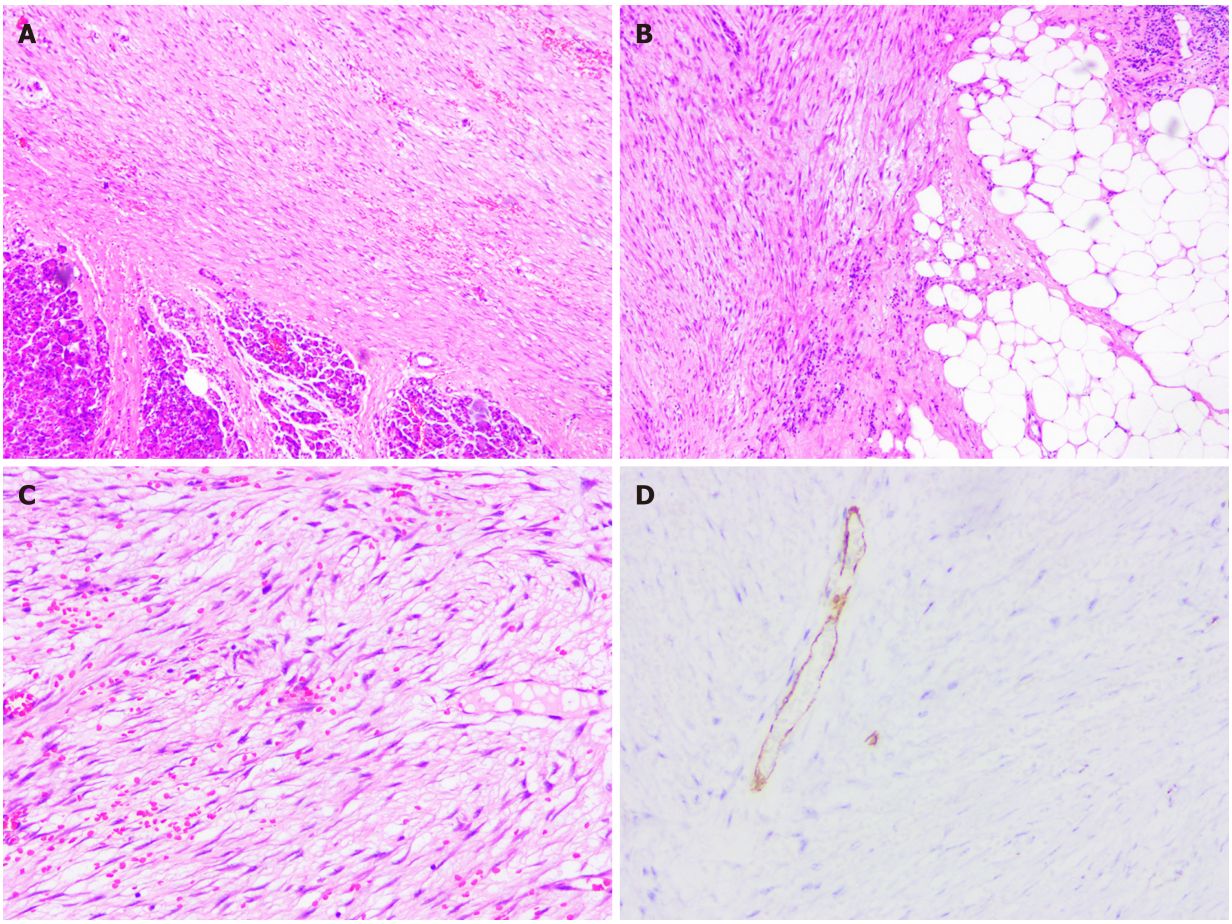

Figure 2 The pathological findings of the resected specimen revealed inflammatory myofibroblastic tumor. A-C: Histological images showed mixed components of dense myofibroblastic tissues and few inflammatory cells, with neoplastic cells infiltrating the surrounding fat tissue (hematoxylin and eosin staining); D: Immunohistochemical studies showed positivity for smooth muscle actin.

pancreatic IMT was more common in the pancreatic head.

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

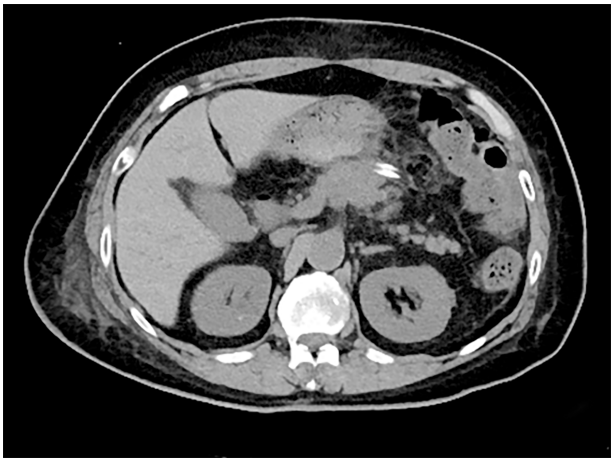

Figure 3 Ten days after surgical resection, computed tomography showed that the pancreatic neck inflammatory myofibroblastic tumor was enucleated, and the tissue of the pancreas remained intact.

the pancreas, and she had no special symptoms.

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.

ACKNOWLEDGEMENTS

The authors would like to thank the support of the Department of Radiology at Shulan (Hangzhou) Hospital Affiliated to Zhejiang Shuren University Shulan International Medical College.

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