



## CASE REPORT

# Recurrence of inflammatory pseudotumor in the distal bile duct: Lessons learned from a single case and reported cases

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## Abstract

Inflammatory myofibroblastic tumors (IMTs) or inflammatory pseudotumors (IPs) have been extensively discussed in the literature. They are usually found in the lung and upper respiratory tract. However, reporting of cases involving the biliopancreatic region has increased over recent years. Immunohistochemical study of these lesions limited to the pancreatic head or distal bile duct seems to be compatible with those observed in a new entity called autoimmune pancreatitis, but usually intense fibrotic reaction (zonation) predominates producing a mass. When this condition is limited to the pancreatic head, the common bile duct might be involved by the inflammatory process and jaundice may occur often resembling adenocarcinoma of the pancreas. We have previously reported a case of IMT arising from the bile duct associated with autoimmune pancreatitis which is an extremely rare entity. Four years after Kaush-Whipple resection, radiological examination on routine follow-up revealed a tumor mass, suggesting local recurrence. Ultrasound-guided FNA confirmed our suspicious diagnosis. This present case, as others, suggests that persistent follow-up is necessary in order to prevent irreversible liver damage at this specific location.

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**Key words:** Inflammatory myofibroblastic tumor; Inflammatory pseudotumor; Local recurrence; Pancreas resection; Whipple procedure

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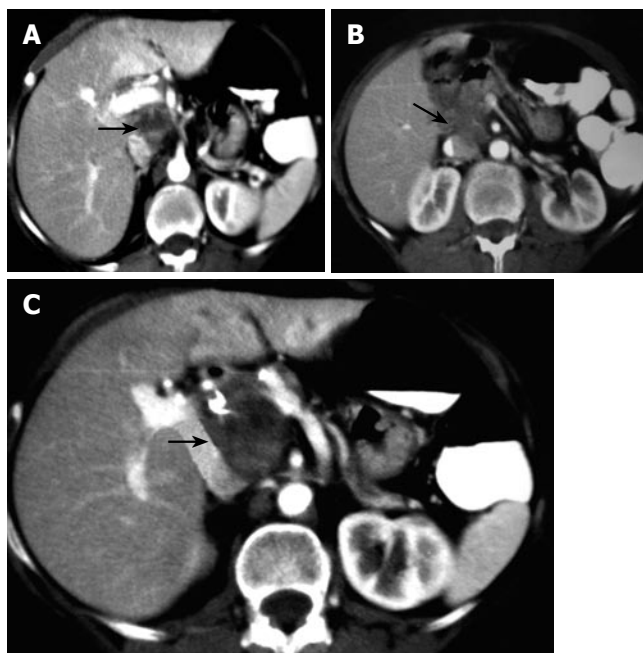
## INTRODUCTION

Histologically, inflammatory myofibroblastic tumor (IMT) is characterized by a proliferation of spindle cells admixed with variable amounts of a lymphoplasmacytic infiltrate. These changes in the pancreatic head produce a mass effect in the biliopancreatic region mimicking a malignant tumor. Thus, surgical resection has been often necessary in order to obtain a definitive diagnosis as well as to relieve symptoms<sup>[1]</sup>. IMT has been shown to possess potentiality for local infiltration and recurrence with persistent local growth at any organ. Hence, a strict follow-up after surgery is necessary because local recurrence may occur many years later. We report herein a case of local IMT recurrence four years after Kausch-Whipple resection with tumor-free margins in a 55-year-old woman. Primary tumor was located in the distal common bile duct accompanied with lymphoplasmacytic reaction in the pancreatic head. To our best of knowledge, this is the first case reporting a local reappearance of inflammatory pseudotumor in the distal bile duct associated with benign lymphoplasmacytic sclerosing pancreatitis that responded successfully to corticosteroid therapy. Further follow-up in the present case is necessary to elucidate if it behaves as a reactive progressive lesion or tumor.

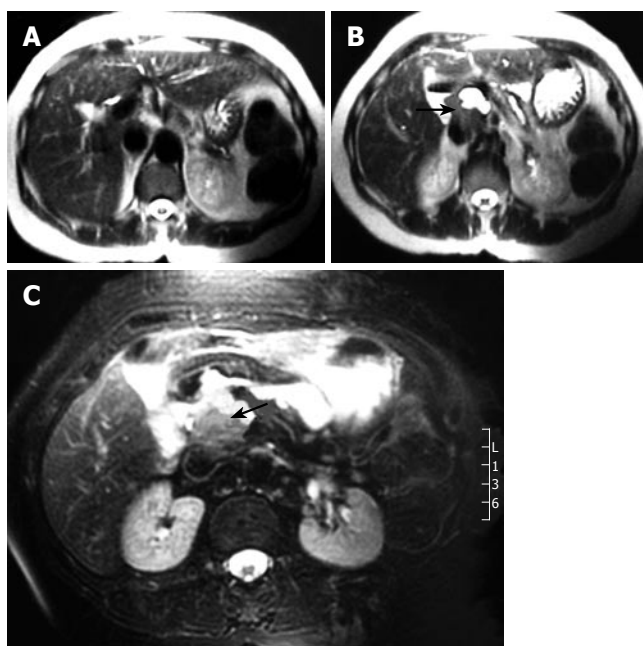
## CASE REPORT

A 55-year-old woman with a past history of IMT affecting the biliopancreatic region [myofibroblastic inflammatory tumor of the distal bile duct (OCD-O code C-24.O M-8825/1) associated with lymphoplasmacytic sclerosing pancreatitis, chronic cholecystitis and reactive lymphadenitis of the isolated nodes] was on routine follow-up after Kaush-Whipple resection.

Four years later, routine surveillance CT scans detected a well defined mass in close relation to the portal vein without apparent infiltration of surrounded structures. At this time, weight loss, malaise or anemia were not present. The tumor mass was well circumscribed, measuring 5 cm × 5 cm, had solid pattern and did not enhance with contrast (Figure 1). An abdominal MRI further delineated that the

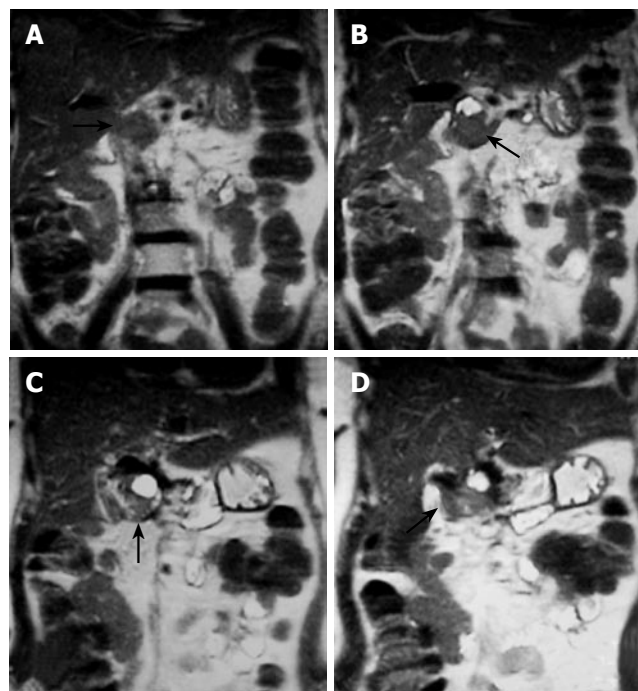


**Figure 1** CT scan revealing a well delimited solid mass (arrow), without enlarged regional or para aortic lymph nodes and any evidence of invasion of other organs.



**Figure 2** CT scan showing a clear relationship between tumor and inferior vena cava vein (arrow).

mass was located between the portal vein and inferior vena cava with no signs of widespread infiltration of organs, thereby strongly suggesting the diagnosis of local recurrence (Figure 2). It did not reveal intrahepatic bile duct dilatation but did show chronic changes in pancreatic tissue (Figure 3). Ultrasound-guided fine-needle aspiration (FNA) was performed for cytological examination, which revealed prominent infiltrate of lymphocytes, plasma cells, and acute inflammatory cells without cytologic atypia. These findings were compatible with IMT. Therefore,



**Figure 3** Serial magnetic resonance imaging of the abdomen (coronal plane) showing the tumor mass in the porta hepatis in close relation with the portal vein and biliary tract.

local recurrence showing benign behaviour was suspected. Then, without evidence of systemic disease, corticosteroid therapy was started. A repeat ultrasound revealed complete remission of the tumor at the 3<sup>rd</sup> mo. The steroid dose is being tapered downward gradually, and we might expect complete remission 6 mo later on low-dose steroid maintenance treatment. On the 5<sup>th</sup> mo of therapy, the patient remains well and liver function tests were within normal limits.

## DISCUSSION

Although IMTs generally behave as benign lesions that are usually cured by radical excision, some cases demonstrate an aggressive growth behaviour. In a study conducted by Coffin *et al*<sup>[2]</sup>, a significant recurrence rate of 25% was demonstrated, likely related to factors precluding complete surgical resection of the pseudotumor. There is a tendency to believe that IMT occurring in the abdomen or retroperitoneum has a propensity for more aggressive behavior with multiple recurrences, invasion into adjacent structures and metastases as shown by DiFiore *et al*<sup>[3]</sup> and Meis *et al*<sup>[4]</sup>. However, other recent reports showed that aggressive behaviour, rapid infiltration and multiple recurrences of IMTs are not limited to abdominal location<sup>[5-7]</sup>. Nowadays, limited evidences exist on the outcomes of patients with a local recurrence of IMT from the biliopancreatic region.

A recent review<sup>[1]</sup> mentioned reported cases of IMT at the biliopancreatic region, describing locoregional recurrences of four cases at the biliopancreatic region<sup>[8,9]</sup> after primary resection of the tumor. Further study of these cases showed that just only one of the four cases represented a true local recurrence of pseudotumor arising

from the bile duct. The other three cases were much different from ours because primary tumor mass was located in the pancreas or because the recurrence was found at a distant site (lung) or the recurrence occurred after a long time interval (17 years) from resection and at a different site from primary tumor. The case reported by Johnson *et al*<sup>[9]</sup> in a 29-year-old black woman recurred after Kaush-Whipple procedure 8 mo later at original site, although the tumor mass was primarily located in the pancreas tissue. Walsh *et al*<sup>[8]</sup> reported three cases of IMT affecting the biliopancreatic region with long-term follow-up. In one of them, recurrence was found at a distant site (lung). In other case, there was a long interval from resection to recurrence in the pancreatic tail, reason why is not considered a true recurrence. If these recurrences represent a second tumor, multicentricity or metastasis is unknown. In the third case, the tumor recurred at the original site (right main hepatic duct) 14 years later, extending into the liver and showed a benign behaviour. This latter case requires further attention as it is similar to ours but has some interesting differences.

It is widely believed today that IMT at the biliopancreatic region, specially distal bile duct, are usually associated with a new entity called autoimmune pancreatitis which is believed to be an initial activation process. There are clear evidences in literature that most of the reported cases of IMT from the distal bile duct showed association with sclerosing lymphoplasmacytic pancreatitis or autoimmune pancreatitis<sup>[1,10-13]</sup> and indicates that the fibroblastic cells in both lesions may be induced at the same site and are usually the same inflammatory and fibrosing process with different histological appearances. However, Walsh *et al*<sup>[8]</sup> did not mention whether the primary tumor at the common bile duct was associated with any form of pancreatitis. Pancreatic tissue was not studied, even they did not mention whether the distal margin was free of inflammatory process. Whether this case represents a form of cholangitis associated with lymphoplasmacytic sclerosing pancreatitis is unknown.

Thus, the case reported by Walsh *et al*<sup>[8]</sup> that did not show any association with autoimmune process is different from ours in which the distal bile duct and pancreatic head was involved by the same inflammatory process. Other important point is that final post-mortem study of the specimen from the case of Walsh *et al*<sup>[8]</sup> showed involvement of the liver which suggests and reinforces previous theories that there is possibly an idiopathic pancreatobiliary inflammatory disease complex involving various processes such as autoimmune pancreatitis, although this was not demonstrated.

Apart from this reported cases, our case represent a true local recurrence of pseudotumor of the distal bile duct associated with autoimmune process in the pancreas head. The present case confirms that IMTs have a tendency to recur with time even after radical excision of primary tumor with free margins. These findings support the theory that IMTs are considered as a systemic disease with different pattern of behaviour in which persistent autoimmune reaction process and multicentricity might be involved<sup>[13,14]</sup>.

Our case seem to accord with the theories previously

developed by Kawaguchi<sup>[15]</sup>, Zen<sup>[13,16]</sup> and Klöppel<sup>[17]</sup> which associates autoimmune pancreatitis with variable involvement of the pancreas and the biliary tract. Fibrosis and zonation<sup>[2]</sup> may occur as a consequence of persistent inflammatory autoimmune process and biliary stenosis might developed in two different ways: due to external compression from fibrotic changes in the pancreatic head and secondary to zonation of the inflammatory process in the biliary tract which represents our case.

This report provides many important messages. First, local recurrence of IMT at biliopancreatic region has been rarely described. To our knowledge, no cases have been reported in the modern era, which usually associate pseudotumor formation in the distal bile duct with a new entity called autoimmune pancreatitis representing zonation of the same inflammatory process. Second, the biologic potential of inflammatory pseudotumors is highly variable (depending on its location and likely related to factors precluding complete surgical resection, such as adherence to vital structures and multifocality), but it generally has an innocuous course, with more local recurrences than distant metastasis as in our case.

Recurrence is common and the use of corticosteroids is permitted. In these cases, as shown by Voss *et al*<sup>[18]</sup> and our present case, local recurrences and systemic disease might not require surgery. However, surgery cannot be excluded and long-term follow-up is necessary in order to prevent irreversible liver damage in this specific location. Usually, acute lesions typically respond to high doses of corticosteroid<sup>[10]</sup>, but chronic lesions such as recurrences, which tend to have more fibrosis, might not respond to medical therapy. Third, our case seems to reinforce the hypothesis that inflammatory myofibroblastic tumors from the bile duct might represent zonation by proximity of an autoimmune process affecting the pancreas. This might be explained by pattern of recurrence: our patient presented 4 years ago with indolent jaundice accompanied by biliary dilatation by inflammatory pseudotumor of the distal bile duct. She underwent Whipple procedure with tumor-free margins and negative isolated nodes. However, 4 years later, remaining asymptomatic, radiologic examinations revealed a tumor mass close to the primary lesion without bile duct compression. Moreover, MRI, liver function test and bilirubin levels were normal. As Whipple procedure separates bile duct from pancreas tissue by interposed bowel loop, where did this recurrence come from? What does the recurrence signify? Perhaps this might be explained by new pseudotumor formation mediated by autoimmune process against remaining pancreatic tissue. Obviously, multicentricity or a new second tumor cannot be excluded.

Many questions still arise about the increasing appearance of inflammatory pseudotumors at different sites: What does really represent inflammatory myofibroblastic tumor? High concentrations of serum Ig subtype, such as IgG4, in IMTs suggest that immunoglobulins may have a pathological role in the fibrosing process mediated by an autoimmune process. Broad pattern of presentation and its ample relation with other autoimmune diseases reinforce this theory<sup>[13,16,19-21]</sup>.

Among other hypothesis, this theory is being increas-

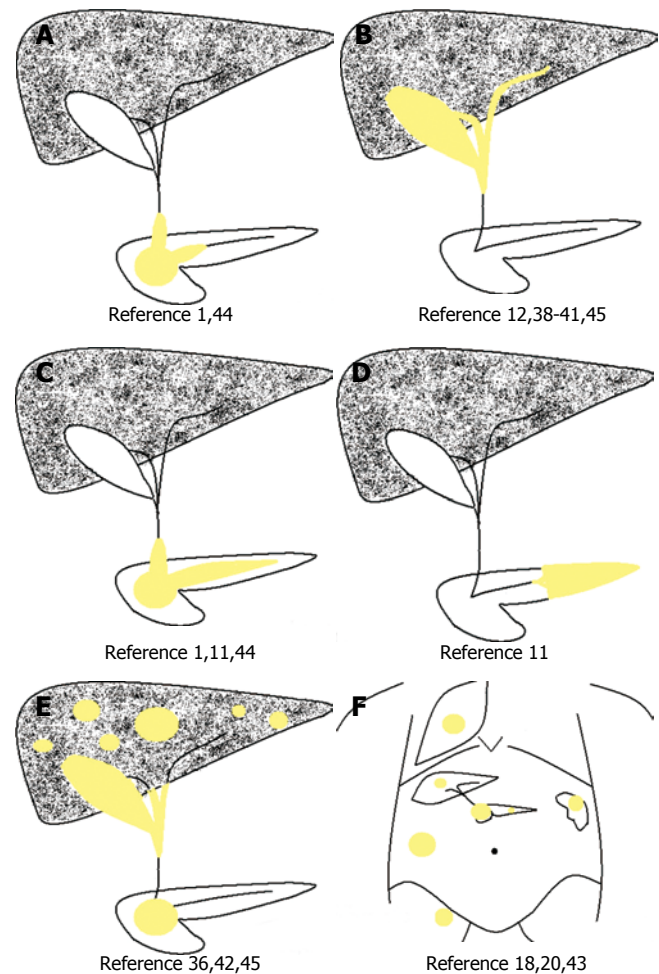


ingly recognized and reflects that inflammatory pseudotumors might represent an advanced stage of an autoimmune process in which intense fibrotic reaction (zonation) predominates<sup>[2,17]</sup>. What evidences exist in favour of the theory that IMTs at the biliopancreatic region are secondary to autoimmune process in the pancreas? Possible evidences are the frequent occurrence of various autoimmune antibodies, such as antibodies against carboanhydrase II and nuclear antigens<sup>[22]</sup>, the elevated IgG4 serum levels and IgG4 positive plasma cells<sup>[13,16,20,23]</sup>, the oligoclonal pattern of T cell receptor gamma gene rearrangements<sup>[24]</sup>, its association with reactive hyperplastic lymphadenopathy<sup>[1,8,25,26]</sup>, and the responsiveness to steroid therapy of the reported cases<sup>[10,27-29]</sup>. More importantly, immunohistochemical results of these tumors are compatible with those of other autoimmune diseases. Is it a systemic disease when found at the biliopancreatic region whose course is determined by its biological activity? Strong association between these tumors and other autoimmune processes suggest that IMTs are advanced reactive lesions secondary to an unidentified agent. High concentrations of serum IgG4 found in reported cases at various sites supports that IMTs represent a systemic condition. Hence, it may be localized, multicentric or systemic at the time of presentation and has a strong tendency to recur. When found at the biliopancreatic region it may represent more than just a biliopancreatic disease.

Some evidences exist about its different biological behaviours that may represent a premalignant state<sup>[2,4,5,29-31]</sup>. What is the pattern of behaviour of inflammatory pseudotumors at the biliopancreatic region? Limited evidence exists to respond this question, but usually behaves as a benign lesion as reported in the literature to date. Our case represents a local recurrence or multicentricity of IMT at the biliopancreatic region that responds well to conservative treatment. A series of four patients previously described by Walsh *et al*<sup>[8]</sup> and Voss *et al*<sup>[18]</sup> demonstrated that IMT from the biliopancreatic region showed a tendency to progressive involvement of multiple organ systems by the same histopathologic process at follow-up.

Is it possible to diagnose preoperatively an inflammatory myofibroblastic tumor presenting with jaundice at the biliopancreatic region and then avoid a complex surgical procedure? A high index of suspicion is necessary to diagnose preoperatively this kind of tumor and exclude malignancy. Medical past history relating other autoimmune process or previously diagnosed inflammatory pseudotumors at any site should be investigated. FNA with immunohistochemical study, and a good response to steroid therapy is necessary to avoid a surgical procedure. Distinguishing inflammatory pseudotumors from malignancy is extremely difficult to do on the basis of radiographic, ultrasound and MRI findings<sup>[1,33]</sup>, specially at the biliopancreatic region as shown by Venkataraman *et al*<sup>[34]</sup>. Agrons *et al*<sup>[35]</sup> agreed with them but referring to extra-abdominal sites (lung). The heightened awareness should prompt more extensive non-invasive and minimally invasive radiologic, histopathologic, and immunohistochemical evaluations before directing patients toward complicated and potentially high-risk surgical procedures.

Further investigation will improve our understanding



**Figure 4** Dynamic classifications for autoimmune pancreatitis. **A-E:** Classification of the variable involvement of the hepatobiliopancreatic system which may be observed in patients with autoimmune pancreatitis. In yellow color is shown the affected zone. <sup>Ref</sup> References of the most relevant and reliable studies that identify this specific pattern of disease. **F:** Multisystemic organ involvement by inflammatory pseudotumor at the biliopancreatic region. In yellow color is shown the affected zone. <sup>Ref</sup> References of the most relevant and reliable studies that identify this specific pattern of disease.

of pathophysiology and etiology of these lesions and such data will help us better recognize this entity preoperatively and differentiate it from carcinomas in order to avoid a surgical procedure. A well illustrated case was the one described by Sasahira *et al*<sup>[36]</sup>.

How should we follow-up these patients? Most organ systems may be involved by primary inflammatory pseudotumors; but also progressive multisystemic organ at follow-up has been described (Figure 4F)<sup>[18]</sup>. It also has been demonstrated that these chronic process may represent an indolent premalignant state<sup>[5,30,31]</sup>. Therefore, it is important to follow-up these patients to rapidly diagnose local or multisystemic recurrences in order to avoid complications specifically at the the biliopancreatic region where these compressing lesions might cause liver damage, cholangitis or pancreatitis. Follow-up must not be limited to the biliopancreatic region but also to other systemic organs. Increase rate in specific IgG4 might indicate reappearance of the autoimmune disease and become a useful marker to detect pseudotumor recurrence<sup>[37]</sup>, but this issue remains

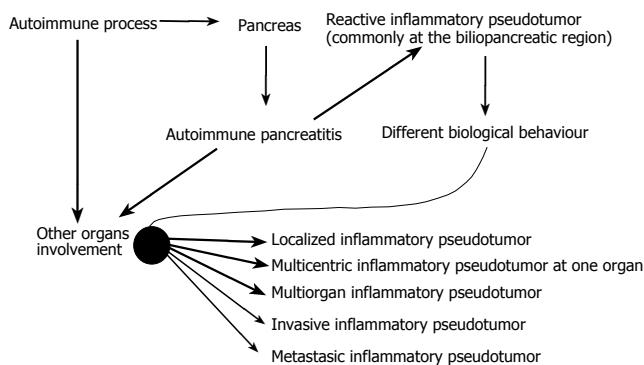


Figure 5 Autoimmune pancreatitis process.

unresolved and controversial<sup>[36]</sup>.

The recurrence in our case is, in fact, represented by multicentricity of the primary tumor, a local recurrence or even systemic disease mediated by an autoimmune process. Overall, we must emphasize that such lesions respond well to corticosteroid therapy and high index of suspicion avoids a surgical procedure.

In conclusion, it seems to be only a question of time until inflammatory pseudotumor will reappear at any site once diagnosed. These lesions most likely will not appear until the etiology is more clearly defined and thus further studies should make a greater emphasis on etiologic factors rather than its presentation form, as virtually any organ may be involved.

Steroid therapy should be the treatment of choice in recurrent cases as good responses have been found in the present and other cases. Follow-up is necessary to avoid complications from recurrences at the biliopancreatic region, especially to avoid liver damage as interval from silent cirrhosis to the development of complication may vary widely. We proposed a new dynamic classification for autoimmune pancreatic-related IMTs<sup>[17]</sup>, with special attention at the biliopancreatic region (Figure 4)<sup>[11,12,18,20,36,38-44]</sup>. Among other theories<sup>[45-51]</sup>, the most widely accepted theory today is that IMT is an auto-immune disorder (Figure 5). IgG4 can be useful as a serological marker of autoimmune pancreatitis and seems also to be useful for the follow-up of patients treated with steroids.

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