Pictorial Essay

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Overlooked and Challenging Encounters–Inflammatory Pseudotumors in the Abdomen and Pelvis: A Pictorial Essay

놓치기 쉽고 진단이 어려운 복부골반강의 염증성 가성 종양: 임상화보

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Inflammatory pseudotumors (IPTs) are uncommon, mass-forming lesions, predominantly involving the lung and orbit. Although the incidence of IPTs is rare in the abdomen and pelvis, they can be encountered as enhancing, soft-tissue lesions, mimicking malignancy or fibrosclerosing disease. Generally, they exhibit a wide range of nonspecific imaging features in various organs. Preoperative imaging diagnosis of IPTs in appropriate clinical settings may help determine proper patient management. In this article, we review radiologic findings of IPTs in the abdominopelvic cavity, including the liver, spleen, kidney, gastrointestinal tract, mesentery, pelvis, and retroperitoneum.

Index terms Inflammatory Pseudotumor; Abdomen; Pelvis; Computed Tomography, X-Ray; Magnetic Resonance Imaging

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INTRODUCTION

Inflammatory pseudotumors (IPTs) are a rare disease entity, commonly involving the lung and orbit but also nearly the whole body (1). In the abdominopelvic cavity, IPTs appear in the liver, spleen, genitourinary tract, gastrointestinal (GI) tract, mesentery,

omentum, and retroperitoneum (2). IPTs are an uncertain process, thought to stand between chronic inflammatory and neoplastic conditions. The disease is also known as various other terms, including plasma cell granuloma, inflammatory myofibroblastic tumor, inflammatory myofibrohistiocytic proliferation, fibrous histiocytoma, xanthoma, xanthogranuloma, fibrous xanthoma, xanthomatouspseudotumor, pseudolymphoma, plasma cell–histiocytoma complex, plasmacytoma, solitary mast cell granuloma, and inflammatory fibrosarcoma (3, 4).

IPTs arise mostly in children and adolescents but may also occur in older patients (4, 5). The cause of IPT remains unknown, and the disease is found in the background of infection, autoimmune disease, trauma, surgery, and malignancy (3, 5, 6). In terms of infection, many microorganisms are implicated for pseudotumors, including *Mycoplasma* and *Nocardia* in the lung, *Escherichia coli, Staphylococcus aureus,* and *actinomycetes* in the liver, and the Epstein-Barr virus in the splenic/nodal disease (3, 7-9). Recently, an association between IPT and immunoglobulin G4 (IgG4)-related sclerosing disease has been established (10). Clinical presentation includes fever, weight loss, thrombocytosis, iron deficiency anemia, hypergammaglobulinemia, and mass effect by a large space-occupying lesion (1-4, 6, 11-13). IPTs manifest from a small solitary mass to an extensive extravisceral infiltrating lesion (2).

With an unclear pathophysiology, IPT is postulated to range from an inflammatory condition to a low-grade malignant process. On histology, IPT demonstrates polymorphous inflammatory cell infiltration, myofibroblastic spindle cells with variable amounts of fibrosis, necrosis, and granulomatous reaction (14-16). It is characterized by histologic variability and complexity. Hussong et al. (17) reported that if IPT shows ganglion-like cells, p53 expression, and aneuploidy, a more aggressive course should be predicted. Anaplastic lymphoma kinase (ALK) overexpression with cytogenic clonal abnormality occurs in approximately 50% of cases of IPT, and ALK positivity is associated with a higher local recurrence and increased fatality (18, 19).

Zen et al. (20) reported that the hepatic IPT is classified into two types: fibrohistiocytic and lymphoplasmacytic. Fibrohistiocytic IPT demonstrates xanthogranulomatous inflammation, neutrophils, and multinucleated giant cells. It occurs in the periphery of the liver with a mass-forming lesion. Further, lymphoplasmacytic IPT is characterized by diffuse lymphoplasmacytic and eosinophilic infiltrations around the hepatic hilum. Venous occlusion, little inflammation, and cholangitis without periductal fibrosis appear in the fibrohistiocytic type. In contrast, the lymphoplasmacytic type shows obliterative phlebitis and cholangitis with periductal fibrosis. IgG4-positive plasma cells are significantly more prominent in the lymphoplasmacytic type than in the fibrohistiocytic type, suggesting an association between IgG4-related disease and lymphoplasmacytic IPT (20).

IPT may show local recurrence and rarely distant metastasis (1, 2). The reported recurrence rate is approximately 25% (3). There are some cases of spontaneous regression or medical treatment of IPT, but mostly they require surgical removal (1-4, 21). Although a benign entity, IPT tends to mimic malignancy in clinical and radiological aspects (2). If adequate surgical management is applied, its prognosis is favorable (1-4, 22). Given the possibility of local recurrence and metastasis, careful follow-ups are required.

IMAGING FEATURES OF IPT

IPTs demonstrate nonspecific and diverse radiologic findings, lacking typical characteristics, even in the same organ (1, 2). In the abdominopelvic cavity, IPTs can occur as a well-circumscribed soft-tissue mass or diffuse ill-defined infiltrative lesion (3, 23). It reflects a variable amount of cellular infiltration and fibrosis (4). The size and enhancement pattern may show chronological changes according to the dynamic course of the inflammatory process. On ultrasonography, IPT is seen as an ill-defined or a well-circumscribed hypoechoic or hyperechoic lesion, with prominent vascularity on Doppler ultrasound (24). IPTs demonstrate heterogeneous attenuation with early peripheral and delayed central enhancement, indicating fibrosis, on CT (4, 24, 25). The lesion contains a hyperintense inflammatory portion and hypointense fibrotic portion on T2-weighted MRI and shows homogeneous or heterogeneous enhancement after contrast injection (23, 26, 27). IPTs occasionally have internal necrosis, ulceration, and mural infiltration (4). Imaging findings of IPT are often confusing, and a wide range of differential diagnosis, including malignancy and other fibrosclerosing disease, such as sclerosing mesenteritis, abdominal fibromatosis, and retroperitoneal fibrosis, is considered (1-3, 23, 28).

HEPATIC IPT

IPTs in the liver show a higher incidence in Asian countries (29). It most commonly occurs in young adults and shows a male predominance (30). Infections, vascular and autoimmune diseases may be accompanied, and other inflammatory conditions (e.g. appendicitis) have also been reported in combination (31-33). Symptoms include abdominal pain, fever, weight loss, portal hypertension, and biliary obstruction (21, 28, 34, 35).

Hepatic IPTs present with nonspecific findings on imaging studies. They mostly appear as a solitary mass but may appear as multiple lesions (1). Ultrasonography shows a hypoechoic or hyperechoic mass with heterogeneous echotexture (36, 37). Some cases of IPT contain internal septation and cystic components (33, 35). Noncontrast CT exhibits hypo- or isoattenuation compared to the muscle (3). On CT, IPTs demonstrate variable contrast enhancement patterns: heterogeneous, homogeneous, septal, peripheral with delayed central, and nonenhancement (Figs. 1, 2) (12, 29). Early peripheral and central filling enhancements reflect retained contrast media in the fibrotic portion in the delayed phase (21, 34, 35). Larger lesions may depict calcification or central necrosis (3). On MRI, the lesion shows T1 low signal intensity and T2 low or high signal intensity (Figs. 3, 4) (38). Uncommonly, IPTs manifest as periportal soft tissue infiltration with adjacent bile ductal dilatation, showing delayed or persistent enhancement (28, 33, 35).

Although IPT is suspected based on the imaging findings, its resemblance to abscesses or malignant hepatic tumors, such as cholangiocarcinoma, hepatocellular carcinoma, and metastasis often requires tissue confirmation using core needle biopsy (3, 28, 33, 35). Further, IPTs involving the biliary tract can be mistaken for a stricture, recurrent pyogenic cholangitis, and periductal infiltrating cholangiocarcinoma (1, 28). After the pathologic confirmation of IPT, the patient mostly undergoes surgical resection, but conservative treatment with nonsteroidal anti-inflammatory drugs is performed at times with spontaneous regression (1, 29, 30).

Fig. 1. Hepatic inflammatory pseudotumor in a 66-year-old woman who presented with generalized weakness.
A, B. Post-contrast portal (A) and delayed (B) phase axial CT show a multiloculated, hypoattenuated mass in the right hemiliver, mimicking a hepatic abscess.

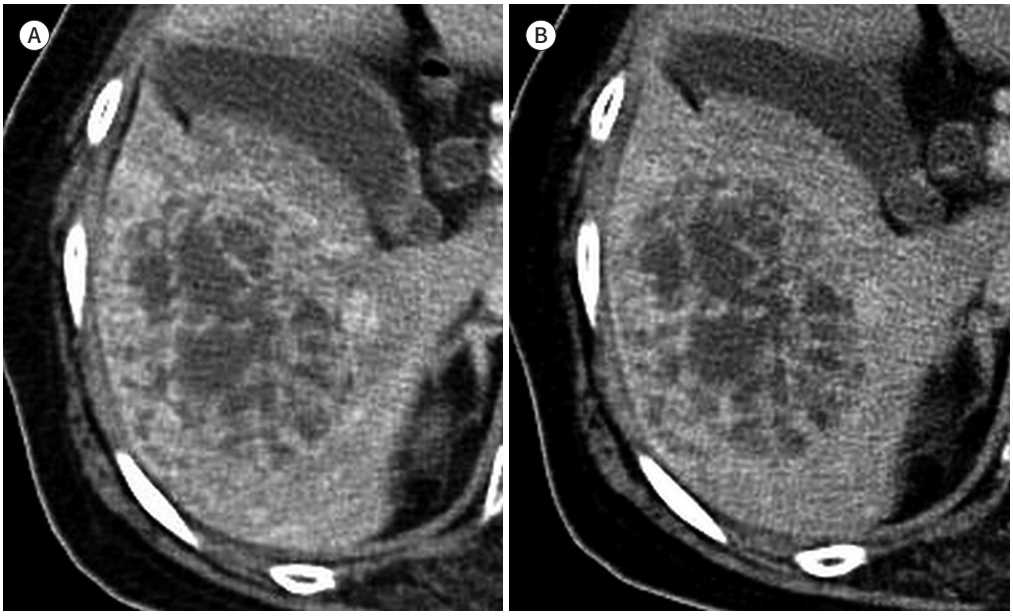
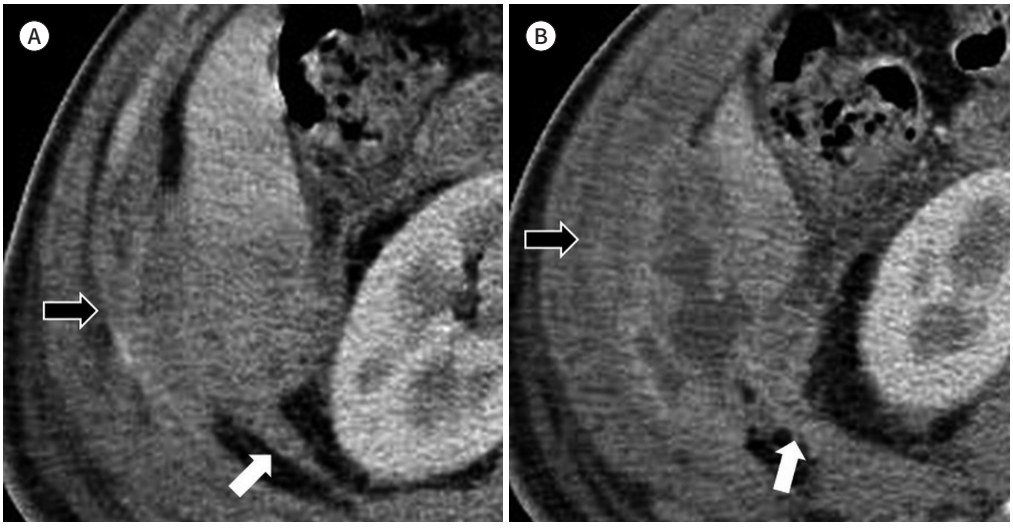


Fig. 2. Hepatic inflammatory pseudotumor in a 63-year-old man who presented with right flank pain.
A, B. Post-contrast CT demonstrates a delayed-enhancing mass with spiculated margins at the tip of the right hemiliver, involving the right renal fascia (white arrows) and the abdominal wall (black arrows).



SPLENIC IPT

There have been a few reports of IPT involving the spleen (1). These cases occurred in middle-aged to old people, with symptoms of weight loss, fever, abdominal pain, and splenomegaly (39, 40). Most cases of splenic IPT manifest as a single well-circumscribed round or oval mass on the radiologic examination (Fig. 5) (2). Varying patterns of calcifications, such as rim-like or stippled shape, may be accompanied (41, 42). The central stellate low attenuation

Fig. 3. Hepatic inflammatory pseudotumor in a 45-year-old man with chronic hepatitis B.
A-F. Pre-contrast T1-weighted MRI (A) shows a well-defined hypointense mass in liver segment 8. The mass depicts enhancement in the early phase (B), washout in the transitional phase (C), and hypointensity in the hepatobiliary phase (D). Fat-suppressed T2-weighted MRI (E) demonstrates intermediate hyperintensity with central hypointensity (arrowheads). Diffusion-weighted imaging with a high b-value (b = 800) (F) shows diffusion restriction. On the basis of a history of chronic hepatitis B, the enhancement profile, and other radiologic findings, the initial diagnosis was hepatocellular carcinoma. After percutaneous core needle biopsy, the mass was confirmed to be an inflammatory pseudotumor.

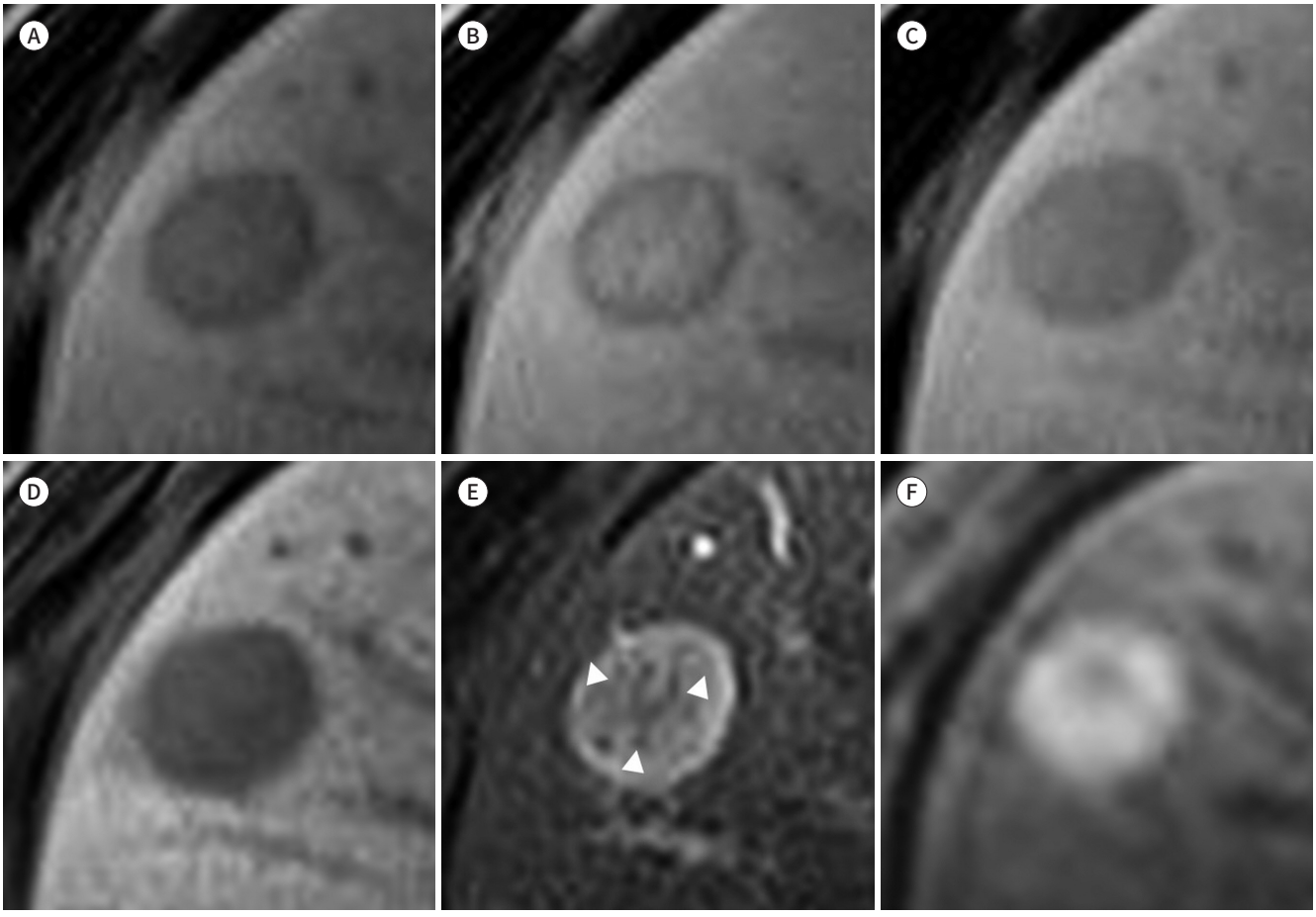


Fig. 4. Hepatic inflammatory pseudotumor in a 49-year-old woman.
A-C. T2-weighted MRI (A) reveals a hypointense mass (white arrow) with internal hyperintense portions (black arrow). In the dynamic portal phase (B), the mass shows hypointensity and extracapsular infiltration (white arrow) in the anterior aspect of liver segment 4. The hepatobiliary phase (C) demonstrates contrast accumulation in the mass.

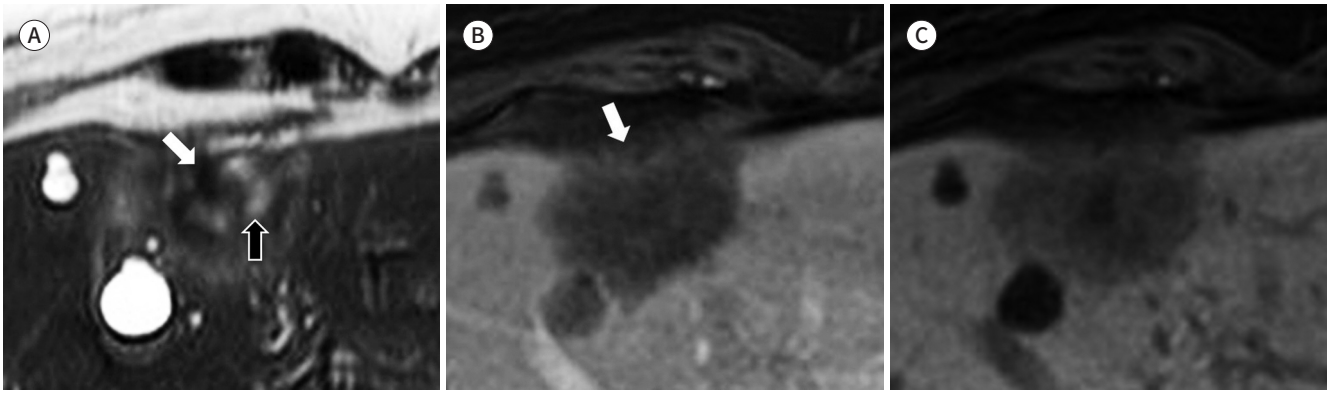


Fig. 5. Splenic inflammatory pseudotumor in a 56-year-old man with a history of colon cancer.
A, B. Post-contrast early (A) and delayed (B) phase CT show a solitary, well-defined mass with delayed enhancement in the spleen, which can be confused with spleen metastasis.

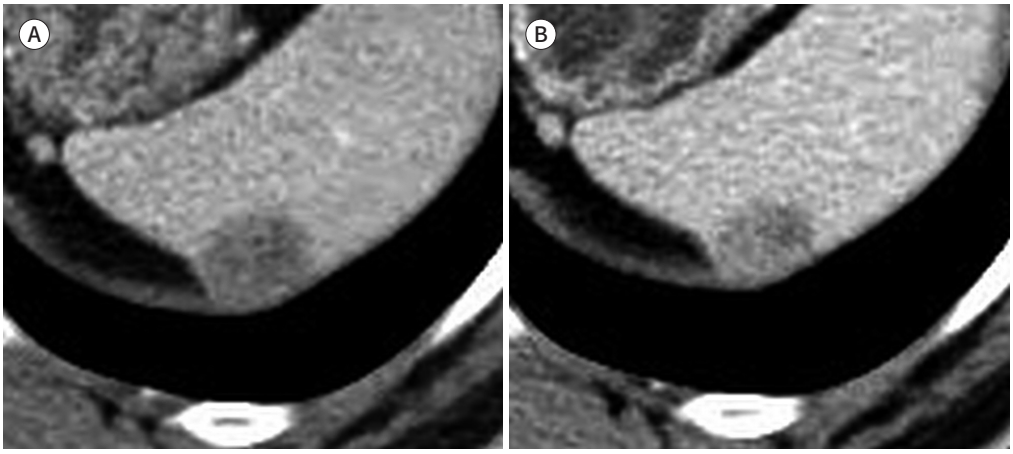
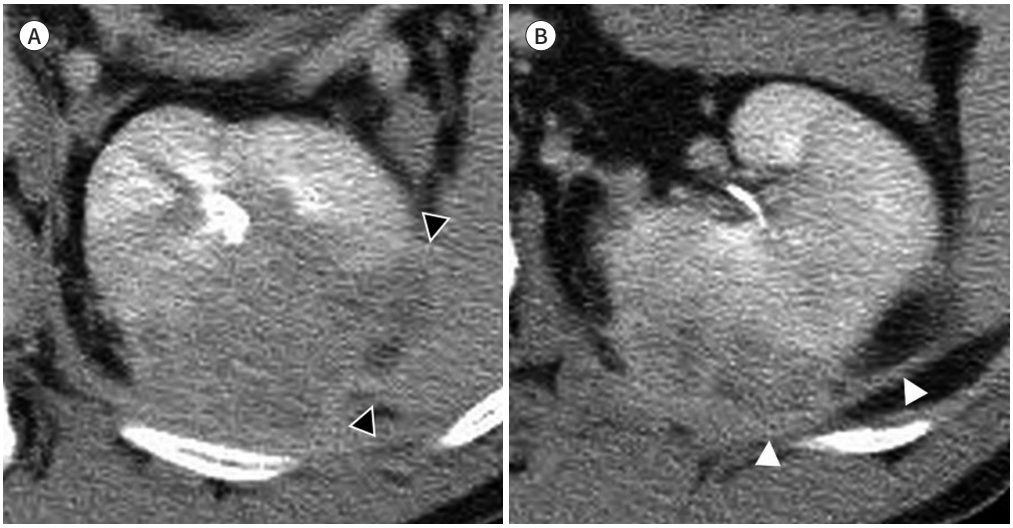


Fig. 6. Renal inflammatory pseudotumor in a 69-year-old man who presented with left flank pain.
A, B. Post-contrast CT demonstrates an inhomogeneously enhancing mass with infiltrative margins in the left kidney. The lesion obliterates the fat plane between the kidney and the spleen (black arrowheads) and involves the left renal fascia (white arrowheads).

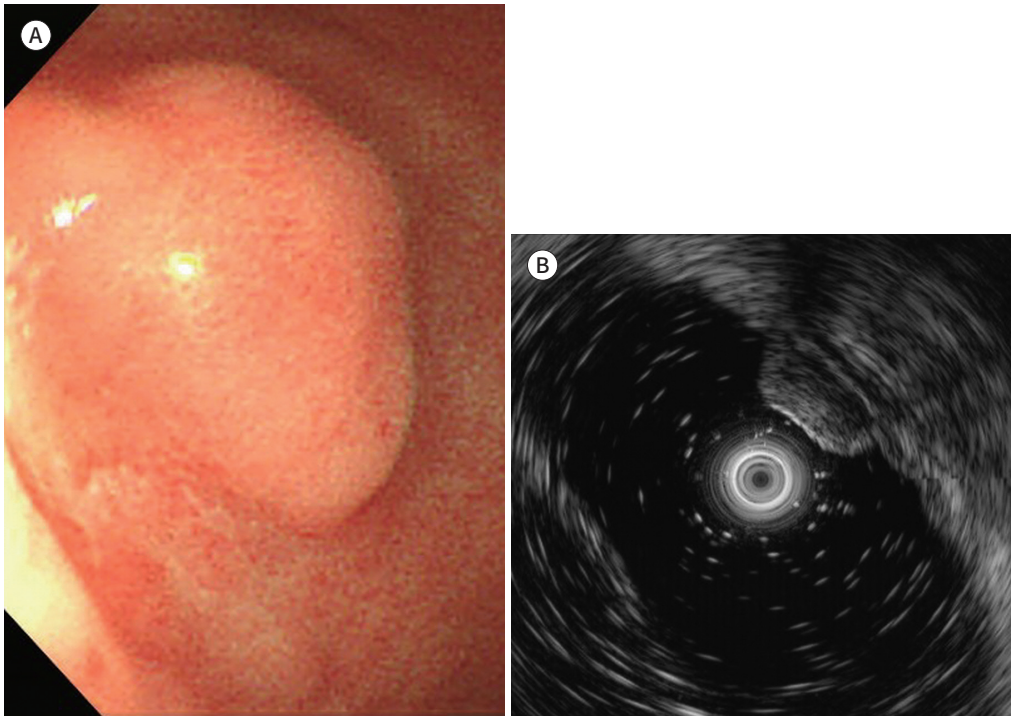


portion, representing fibrosis on CT was documented (41, 42). On MRI, the mass shows iso- or hyperintense T1 signal intensity and hypo- or hyper T2 signal intensity with delayed enhancement (42). Splenic IPTs often mimic lymphoma, metastasis, and other tumors, such as hamartoma (40). However, a favorable prognosis is followed after splenectomy (39).

RENAL IPT

IPTs arising in the kidney are extremely rare (1). Patient's age is variable from children to adults (2). Renal IPTs are more common in men and manifests as fever, flank pain, and hematuria (4). Radiologic findings are noted with either a well-defined or ill-defined heterogeneous or uniformly hypoechoic mass on ultrasound, low attenuation with hyperattenuating foci from calcification on CT, and hypovascular tumor on MRI (Fig. 6) (1, 4). Enhancement

Fig. 7. Stomach inflammatory pseudotumor in a 63-year-old woman.
A, B. Endoscopic image (A) shows a single polypoid subepithelial mass in the anterior wall of the proximal antrum of the stomach. Endoluminal ultrasonography (B) reveals an oval, hypoechoic mass with smooth margins, which is continuous with the second gastric wall layer.



pattern of mass includes corticomedullary phase enhancement and excretory phase washout (2).

Other sites of IPT in the genitourinary tract are the bladder and adrenal gland. Bladder IPTs can be associated with a previous trauma or surgery and presents as a polypoid enhancing intraluminal mass or submucosal mass with or without perivesical fat extension (43). IPT might be considered when an enhancing tumor is surrounded by a clot, particularly in young adults. If bladder IPT occurs in a child, it should be differentiated from rhabdomyosarcoma (43). Further, IPTs are very rare in the adrenal gland, showing a nonspecific adrenal solid mass (4).

GI TRACT IPT

IPTs in the GI tract uncommonly involve the stomach, small bowel, colon, and esophagus (1). Symptoms of GI tract IPT include fever, abdominal pain, dysphagia, bowel obstruction, and anemia (3, 5). It demonstrates a soft tissue mass with luminal narrowing and associated mesenteric change or diffuse infiltrative wall thickening (2). The mass occasionally shows calcification, ulceration, and extramural extension (Fig. 7) (5).

GI IPTs are treated with surgical resection, but conservative treatment with steroids, nonsteroidal anti-inflammatory drugs, and radiation has been reported. Local recurrence is possible with an aggressive pattern, and there is a small chance of malignant transformation (5).

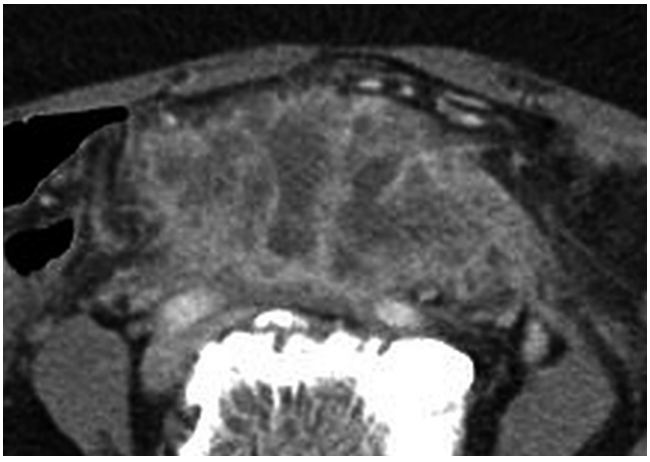


Fig. 8. Mesenteric inflammatory pseudotumor in a 71-year-old woman who presented with abdominal pain.
Axial post-contrast CT demonstrates a heterogeneously enhancing mass with spiculated margins in the small bowel mesentery.

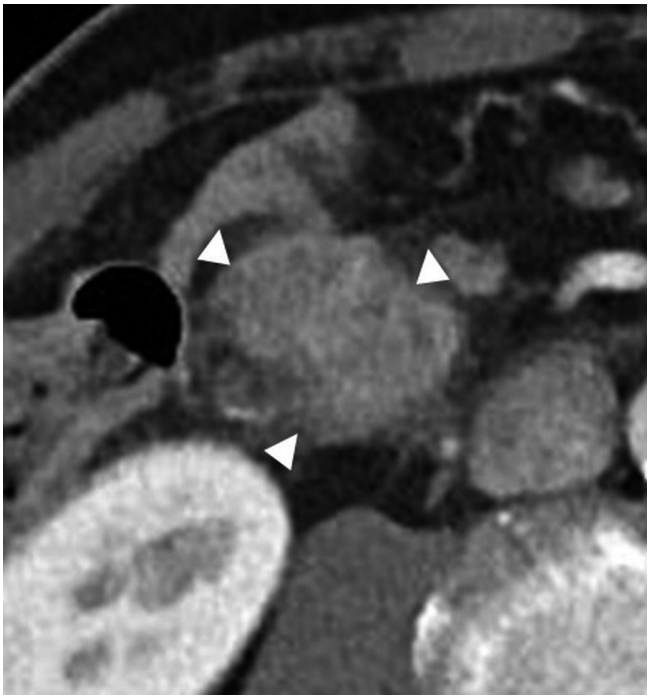


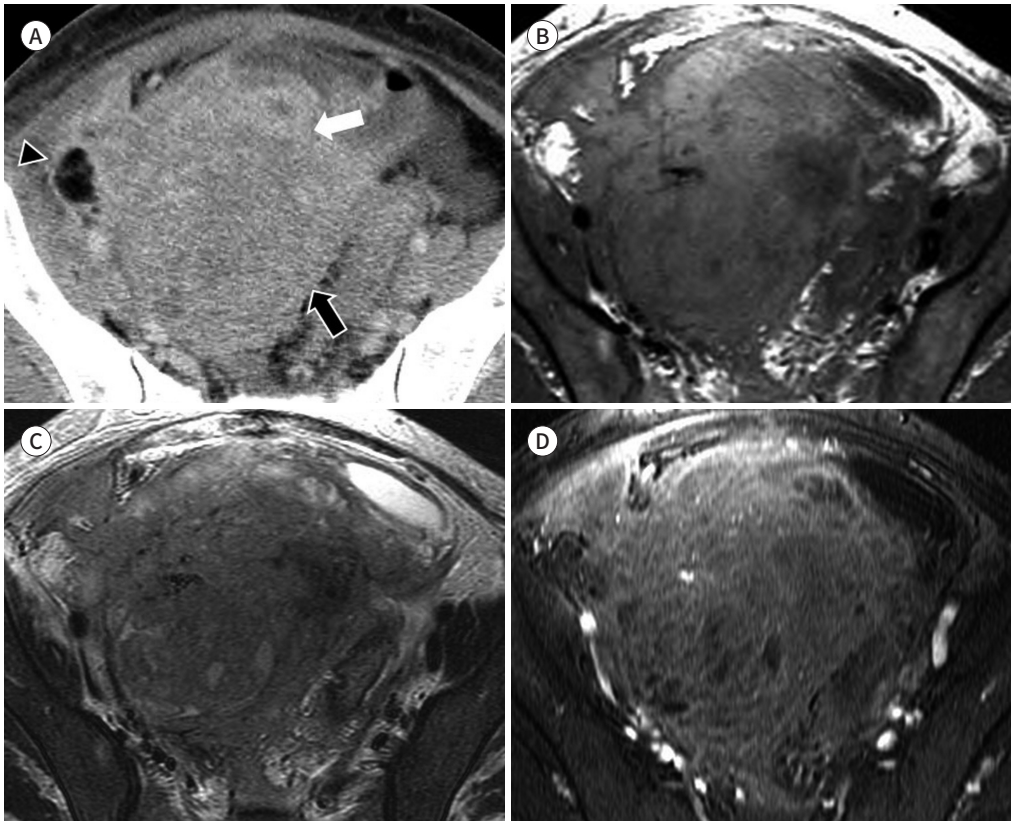
Fig. 9. Mesenteric inflammatory pseudotumor in a 62-year-old man with a history of stomach cancer.
Axial CT shows an enhancing mass (arrowheads) with soft tissue density and partial, ill-defined margins in the mesentery, mimicking a metastatic tumor.

MESENTERIC IPT

IPTs involving the mesentery are uncommon and tend to appear in children and adolescents (1). Patients present with fever, malaise, abdominal pain, and weight loss (1). Mesenteric IPTs may be a well-circumscribed mass or an ill-defined infiltrative lesion with invasion of the adjacent bowel (44, 45). On ultrasound, a well-defined solid, mixed echogenic mass is seen. Enhancement patterns range from nonenhancement to peripheral or heterogeneous enhancement on CT (Figs. 8, 9) (46). IPT might have central necrosis, showing a nonenhancing low-density portion (3, 46). Mesenteric fibromatosis, lymphoma, sarcoma, and metastasis are included in the differential diagnoses (45).

Mesenteric IPTs are cured with complete surgical resection. If the mass does not undergo margin-negative surgical removal, an adjuvant treatment, such as chemotherapy, corticoste-

Fig. 10. Pelvic inflammatory pseudotumor in a 44-year-old woman who presented with abdominal pain.
A-D. Post-contrast CT (A) shows a large, heterogeneously attenuated mass with enhancement in the pelvic cavity, involving the bowel (white arrow), the uterus (black arrow), and the right pelvic wall (arrowhead). Pre-contrast T1-weighted (B) and T2-weighted (C) MRI depict mixed heterogeneous signal intensities of the mass. After contrast enhancement (D), the mass demonstrates strong enhancement.



roids, or anti-inflammatory drugs, is provided, but its success rate is low. Patients with mesenteric IPTs have a recurrence rate of 15–37% and high risk of sarcomatous change (47).

PELVIC AND RETROPERITONEAL IPTS

Pelvic and retroperitoneal IPTs are very rare (2). They demonstrate a large soft tissue mass with heterogeneous or mixed attenuation and homogeneous or heterogeneous enhancement (4, 48). There is an accompanying mass effect on the neighboring structures. On MRI, slightly or markedly high T2 signal intensity and iso- or slightly high T1 signal intensity are depicted (Fig. 10) (2). It shows a massively infiltrative lesion, which mimics malignancy (e.g., sarcoma, lymphoma), infection (e.g., actinomycosis), and retroperitoneal fibrosis (Figs. 11, 12).

CONCLUSION

Radiologic findings of IPT are nonspecific and broad in the abdominopelvic cavity. However, in some cases, IPTs manifest as a soft tissue lesion demonstrating well-defined or infiltrative margins and delayed contrast enhancement, focal T2 hypointensity with variable proportions of fibrotic components. IPTs may be confined in a single organ or extended through

Fig. 11. Pelvic inflammatory pseudotumor in a 49-year-old woman.
A, B. Axial post-contrast CT (A) shows an ill-defined, enhancing lesion in the left aspect of the anterior pelvic cavity. Coronal CT (B) reveals involvement of the urinary bladder (arrows).

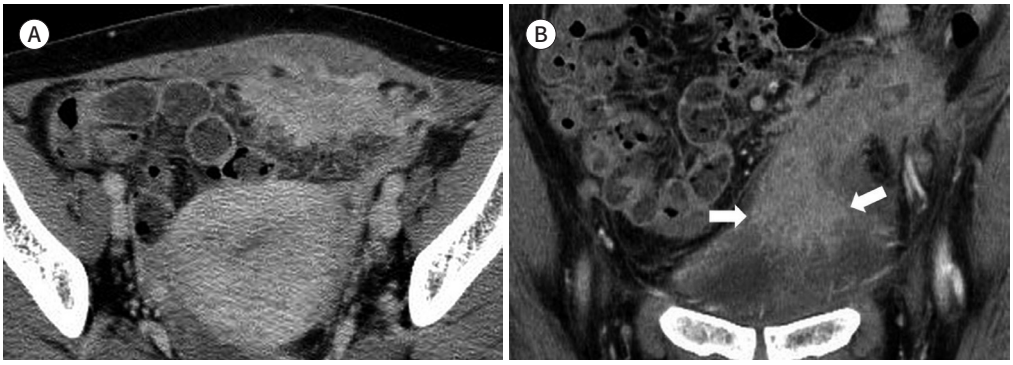
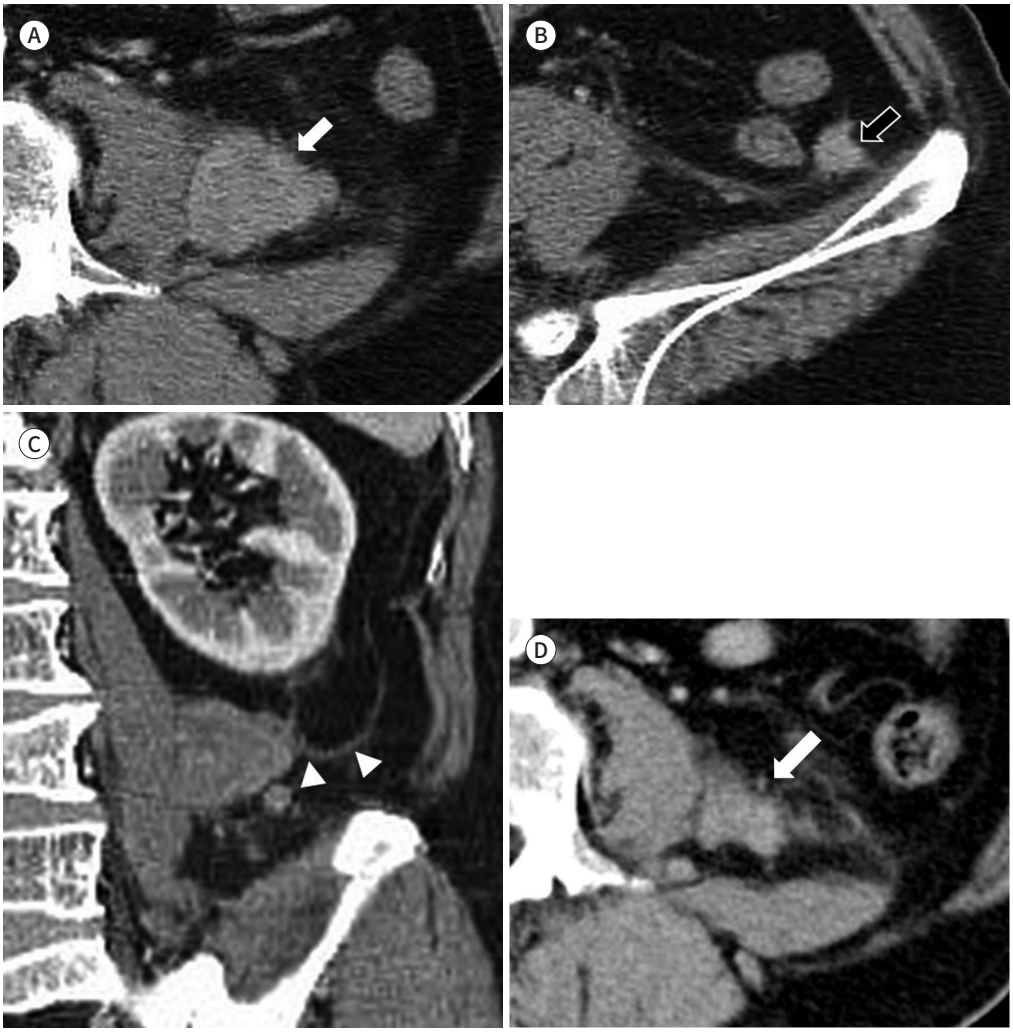


Fig. 12. Retroperitoneal inflammatory pseudotumor in a 74-year-old man.
A-D. Post-contrast CT shows a delayed-enhancing mass with high attenuation in the left retroperitoneum (A, arrow) and another satellite nodule (B, arrow). Coronal CT (C) demonstrates left lateroconal fascial thickening (arrowheads) and perilesional fat stranding. After three months, post-contrast CT reveals a decrease in the size of the retroperitoneal inflammatory pseudotumor without treatment (D, arrow).



the fascial plane to other sites.

Although IPTs are regarded as a benign disease with a mixture of inflammation and myofibroblastic cell proliferation, it resembles a malignant tumor on clinical and radiological context, with a rare possibility of recurrence and metastasis. Accurate imaging diagnosis of IPT is challenging for radiologists, and many tumorous conditions are considered in the differential diagnosis. Particularly, in oncologic patients, tumefactive growth pattern of IPT induces a preceding impression of tumor metastasis or seeding. In our series, eight of twelve cases were surgically removed and four cases were diagnosed with percutaneous core needle biopsy, followed by intensive observation. Appropriate preoperative establishment of the diagnosis may lead to prevention of unnecessary radical surgery, particularly in incidentally found IPT with an oncological background. In conclusion, knowledge of diverse imaging characteristics of IPT helps the radiologist guide proper management and better outcome of patients.

Author Contributions

Conceptualization, P.J.Y., J.H.W., H.J.Y.; data curation, K.M.H., P.J.Y., L.J.H.; formal analysis, K.M.H., P.J.Y.; investigation, K.M.H., P.J.Y.; methodology, P.J.Y., K.Y.S.; project administration, P.J.Y., K.Y.S.; resources, P.J.Y., K.M.H., P.J.W.; supervision, P.J.Y., J.H.W.; validation, P.J.Y., K.Y.S., H.J.Y.; visualization, K.M.H., P.J.Y.; writing—original draft, K.M.H., P.J.Y.; and writing—review & editing, P.J.Y., K.Y.S.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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놓치기 쉽고 진단이 어려운 복부골반강의 염증성 가성 종양: 임상화보

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염증성 가성 종양은 주로 폐와 안와에서 드물게 발생하는 종괴 형성 병변이다. 복부 및 골반에서의 염증성 가성 종양의 발생은 매우 드물지만, 이는 조영증강되는 연부조직 병변으로 나타날 수 있으며 악성 종양 혹은 섬유경화질환과 혼동될 수 있다. 대개 이는 여러 장기에서 다양한 불특정 영상 소견을 보인다. 적절한 임상 정보와 함께 술전 염증성 가성 종양의 진단이 이루어지는 것은 알맞은 환자 처치를 도울 수 있다. 현 임상화보에서는 간, 비장, 신장, 위장관, 장간막, 골반, 후복강에서 발생한 복부골반강의 염증성 가성 종양의 영상의학적 소견을 정리한다.

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