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Case Report

Mesenteric inflammatory pseudotumor: A rare case report and review of the literature ☆,☆☆

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ABSTRACT

Inflammatory pseudotumor (IP) is a rare type of benign tumor. Although initially identified in the lung, it has now been identified in a number of somatic and visceral sites, but mesenteric presentation is uncommon and has a variable clinical presentation. As inflammatory pseudotumor mimic malignancy both clinically and radiologically, the radiologist should be familiar with this entity. The only effective treatment is complete surgical resection. We present the case of a 55-year-old woman who presented with chronic abdominal pain and was diagnosed with a mesenteric inflammatory pseudotumor, in an attempt to illustrate the different imaging aspects of this benign condition in ultrasound, computed tomography and magnetic resonance imaging, and to simplify the description of these tumors.

Introduction

Inflammatory pseudotumor (IP) is a rare benign neoplasm first described by Brunn in 1939 [1]. Most inflammatory pseudotumor of the gastrointestinal tract or mesentery occur in children and young adults. Mesenteric involvement in older adults is extremely rare [2].

Although initially identified in the lung, it has since been described in a number of somatic and visceral regions.

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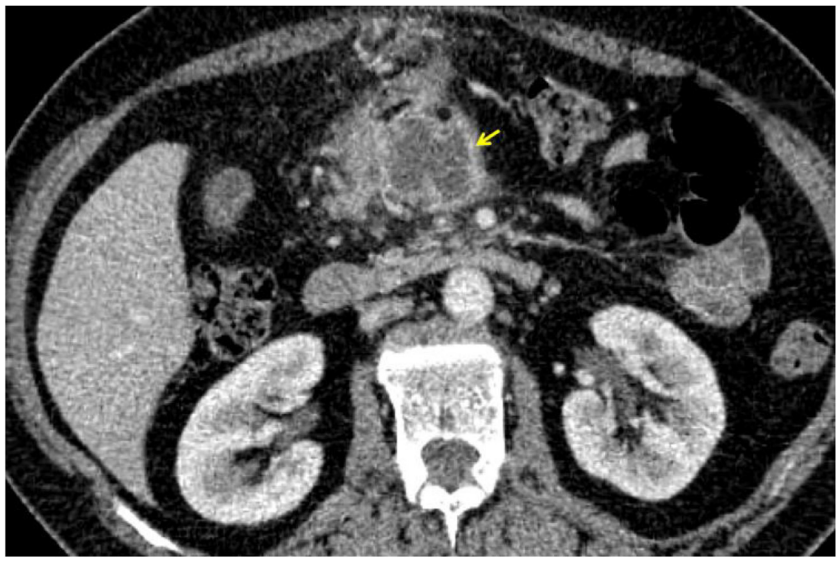

Fig. 1 – Axial enhanced computed tomography of the abdomen showing a large necrotic peritoneal mass (yellow arrow) with infiltration of surrounding fat.

The aetiology of IP is unknown, but likely explanations include previous inflammatory or infectious causes and possibly foreign body reactions. In the majority of cases, no causative agent can be identified [3] . The clinical presentation is nonspecific, with a progressive onset of symptoms including abdominal pain, fever, malaise, weight loss and anemia. Radiological studies can hardly distinguish IPs from other tumors, therefore a histological study is required for diagnosis [4].

We report a pathologically proven case of a mesenteric inflammatory pseudotumor with extensive central necrosis mimicking a necrotized malignant mass in a middle-aged patient.

Case report

A 55-year-old female patient was admitted to hospital with nonspecific abdominal symptoms, including diffuse and intermittent abdominal pain, nausea and anorexia. The symptoms had been present for approximately 2 weeks and were progressively worsening.

On physical examination, the patient was alert and cooperative. Her weight and height were within the normal range for her age. Body temperature was 37°C.

Abdominal examination revealed tenderness to palpation in the epigastric and periumbilical regions without palpable masses. Other systems were unremarkable.

Initial abdominal ultrasound showed a large mass in the perigastric region, and this mass showed a mixed echo structure within the peritoneum.

Contrast-enhanced computed tomography showed a large necrotic peritoneal mass with infiltration of surrounding fat (Fig. 1), adherent to adjacent structures including the stomach, pancreas and transverse colon (Fig. 2). No metastases were found in other abdominal organs.

Consequently, the patient was admitted for surgery, where a well-vascularized mass was found adjacent to the stomach, transverse colon, body of the pancreas and superior mesenteric artery and vein. Further exploration allowed these vessels to be preserved, but a segmental colectomy of the transverse colon and partial gastrectomy were performed. Intraoperative pathological examination was performed and confirmed the absence of malignancy. Definitive histology revealed a mesenteric inflammatory pseudotumor (Fig. 3). The patient was discharged from hospital after 2 weeks. At 1 year follow-up, there was no clinical or imaging evidence of recurrence.

Discussion

Inflammatory pseudotumor are most common in children between 2 and 16 years of age, although adults (as in our case) can also be affected [5]. Retrospective reviews have been published in the literature for the juvenile population, but only small series have been documented for adults, making IP a rare disease [4], with females being slightly more commonly affected than males [6]. IP can affect almost any organ in the human body, although the lungs and orbit are most commonly affected[4]. The most common extrapulmonary sites of IP are the intra-abdominal omentum and mesentery, accounting for 43% of IPs. Other intra-abdominal organs involved include the stomach, liver, gallbladder, colon, spleen, and pancreas [7].

The aetiology and pathogenesis of inflammatory pseudotumor are poorly understood. Initially thought to be reactive, secondary to surgery, minor trauma, occult infection, or associated with systemic disease, they are now classified as neoplasms, especially since studies have identified ALK gene mutations in 50% of cases. This explains the tendency of these lesions to recur and their invasive and metastatic potential [3].

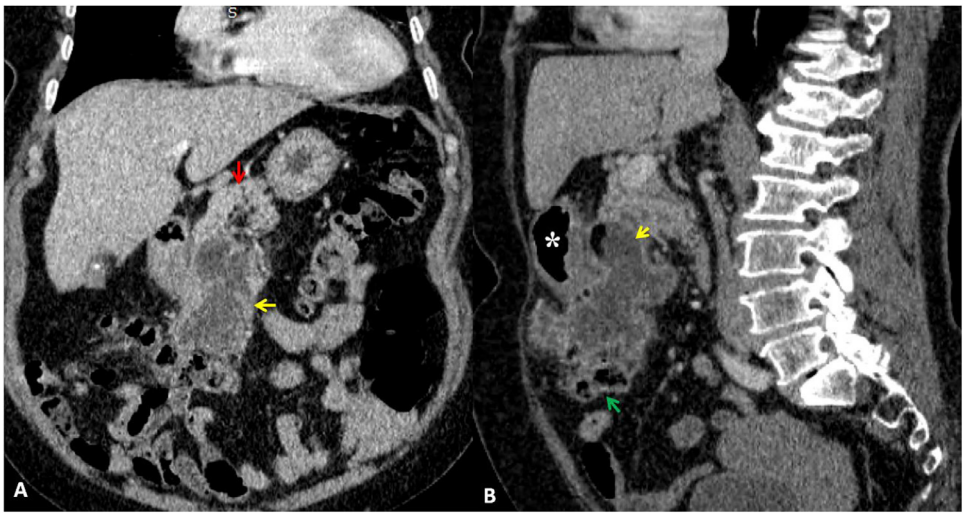

Fig. 2 – Coronal (A) and sagittal (B) enhanced computed tomography of the abdomen showing the necrotic peritoneal mass (yellow arrow) adherent to adjacent structures such as the transverse colon (green arrow), pancreas (red arrow) and the stomach (*).

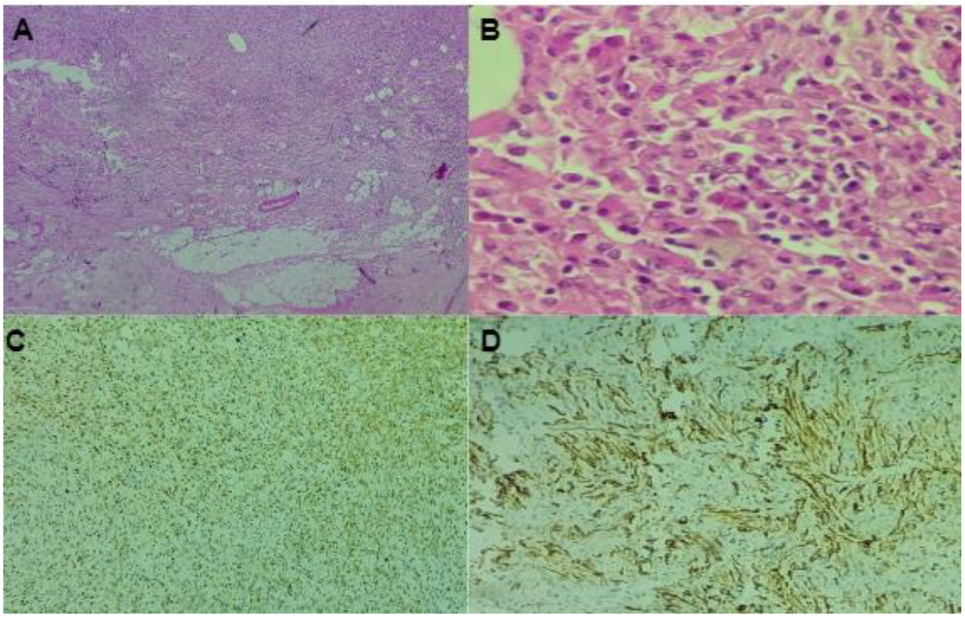

Fig. 3 – Histological and immunohistochemical staining photomicrographs show areas of histiocytes, alternating between foamy and spindle-shaped", associated with numerous myofibroblasts, plasma cells, lymphocytes and a few neutrophils and eosinophils (A, B: H-E staining: original magnification x 40 and 400), cytoplasmic expression of CD68 (C: CD68: original magnification x100), membranous and cytoplasmic expression of SMA (D: SMA: original magnification x 100).

It is a rare histologically benign mass characterized by proliferative myofibroblasts, fibroblasts, histiocytes, plasma cells, and lymphocytes. [2]. These lesions are called pseudotumor because their tendency to invade surrounding tissues mimics the behavior of malignant tumors [8].

Several alternative terms have been used to describe this lesion since its initial report, including postinflammatory pseudotumor, cellular inflammatory pseudotumor, plasma cell pseudotumor, plasma cell granuloma, inflammatory myofibroblastic tumor, inflammatory myohistiocytic proliferation, inflammatory myohistiocytic tumor, inflammatory pseudosarcomatous proliferation, inflammatory fibrosarcoma, myofibroblastoma, fibrous histiocytoma, histiocytoma, solitary xanthoma, xanthogranuloma, fibroxantholoma, omental-mesenteric myxoid hamartoma, pulmonary plasmocytoma, mast cell tumor [3].

The clinical presentation of abdominal IP is often abdominal pain; this may be accompanied by systemic symptoms common to IP at other sites, including weight loss, failure to thrive, fever, anemia, thrombocytosis, and/or hypergammaglobulinemia [3].

Inflammatory mesenteric pseudotumors have nonspecific imaging features. It appears as a solid, mixed echotexture mass within the mesentery that is well defined or infiltrating on sonograms. Doppler ultrasound may show prominent vascularity [9].

On CT scan, there is variability in enhancement characteristics, ranging from heterogeneous or peripheral enhancement to nonenhancement. Central calcifications may be present. Larger tumors have been shown to have low density in the center of the mass, indicating necrosis [10], and the tumors may present as multifocal disease [2].

The appearance of these tumors on MRI images is also variable, typically hypointense relative to skeletal muscle on T1-weighted images, hyperintense on T2-weighted images, and heterogeneously enhanced with contrast if contrast material is administered [1].

The differential diagnosis of inflammatory pseudotumors includes the mesenteric fibromatosis, lymphoma, metastatic disease, and soft tissue sarcomas [9].

Mesenteric inflammatory myofibroblastic tumor may be solitary or multicentric. Our patient with mesenteric tumor had a solitary mass. The solitary lesions may be large, circumscribed and lobular with extensive adhesion to adjacent structures requiring radical excision [5]. While the majority of intra-abdominal myofibroblastic tumors show small areas of necrosis, as in our patient, extensive irregular necrosis may develop within the mass. A necrotized malignant tumor or abscess could be an important differential diagnosis in our case.

Although clinical, radiological, and intraoperative findings are informative, the histology and/or immunohistochemical staining of resected tumors is necessary for the definitive diagnosis of mesenteric IPs [11]. Hematoxylin-eosin is the most commonly used stain for histopathological examination and can identify spindle cells, myofibroblasts, plasma cells, lymphocytes, and histiocytes. The most commonly used immunohistochemical stains are SMA (positivity: 80–90 %), desmin (positivity: 60%-70%), ALK-1 (positivity: 33%-50%), creatine (positivity: 30%), COX-2, and VEGF [12,13].

The differential diagnosis by histopathology should include sarcomatoid carcinoma, desmoid tumor, sclerosing mesenteritis, gastrointestinal stromal tumor, schwannoma, leiomyoma, leiomyosarcoma, solitary fibrous tumor, sarcomatoid mesothelioma, and well-differentiated inflammatory liposarcoma [7].

As IP has a tendency to spread and recur, total resection is the best course of treatment. Treatment recommendations have not yet been agreed upon for cases where surgical resection is not feasible or cannot be completed because of tumor proximity to vital organs, positive surgical margins, or distant metastases. However, chemotherapy, radiotherapy, steroids, anti-inflammatory medications, or tyrosine kinase inhibitors have all been suggested [7].

Following surgical or pharmacological treatment, close CT follow-up is essential for early detection of tumor recurrence.

Conclusion

The diagnosis of mesenteric inflammatory pseudotumor is rarely established prior to surgery due to the nonspecificity of imaging and laboratory tests. Therefore, a mesenteric inflammatory pseudotumor should be considered when a mass presents in an unusual location. Whenever possible, complete surgical resection should be performed and the patient followed up for the long term.

Guarantor of submission

The corresponding author is the guarantor of submission.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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