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Mesenteric inflammatory pseudotumor: A difficult diagnosis. Case report

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ABSTRACT

INTRODUCTION: Inflammatory pseudotumor (IP) is an uncommon benign neoplasm. It was first described in the lung but it has been recognized in several somatic and visceral locations. Mesenteric presentation is rare and its clinical presentation is variable but patients can be completely asymptomatic. Complete surgical resection is the only curable treatment. Rational follow-up protocols have not been established yet.

PRESENTATION OF CASE: A 57 years-old man, with no relevant comorbidities and completely asymptomatic, apart from a lump on the right hypochondrium, was submitted to surgical resection of a large mesenteric mass. The preoperative Computed Tomography suggested gastrointestinal stromal tumor as the most probable diagnosis. Definitive histological examination of the completely resected surgical specimen confirmed the diagnosis of IP. The patient has been on follow-up for four years, without no evidence of recurrence.

DISCUSSION: The preoperative diagnosis of IP may be difficult to establish mainly due to the lack of a typical clinical presentation. It is a rare entity, particularly in the adult population. These two aspects make it easier to neglect this entity in the differential diagnosis of an abdominal mass on asymptomatic adults. Although there are no formal guidelines on follow-up, close follow-up seems to be advisable in these patients as recurrence is frequent.

CONCLUSION: IP should be present as a possible differential diagnosis in an abdominal mass. Complete excision of the lesion can be curable but close follow-up seems to be required.

1. Introduction

The present work has been reported in line with the SCARE criteria [1].

Inflammatory pseudotumour (IP) is a rare benign neoplasm firstly described by Brunn in 1939 [2]. It was first described in the lung but it has been recognized in several somatic and visceral locations [3]. Since its first description, several other terms have been used to address this lesion, such as *postinflammatory pseudotumour*, *cellular inflammatory pseudotumour*, *plasma cell pseudotumour*, *plasma cell granuloma*, *inflammatory myofibroblastic tumour*, *inflammatory myohistiocytic proliferation*, *inflammatory myohistiocytic tumour*, *inflammatory pseudosarcomatous proliferation*, *inflammatory fibrosarcoma*, *myofibroblastoma*, *fibrous histiocytoma*, *histiocytoma*, *solitary xanthoma*, *xanthogranuloma*, *fibroxantholoma*, *omental-mesenteric myxoid hamartoma*, *pulmonary plasmocytoma*, *mast cell tumour* [4].

IP is a lesion composed of inflammatory cells and myofibroblastic spindle cells which can pose, on the preoperative setting, a difficult differential diagnosis with malignacy [2].

Its exact etiology is largely unknown, but it may be related to with minor trauma, surgery, other malignancy or even previous infection [2,5].

Clinical presentation is quite unspecific, as fever, weight loss, malaise and/or abdominal pain may be present. Non-specific laboratory results may be present [3,6].

IPs can be hardly distinguished from other tumors on radiologic exams, making histologic analysis mandatory to establish the diagnosis [7].

Despite its usual benign clinical behavior, IPs can turn out to be aggressive and recurrent [3]. To the best of our knowledge, no definitive guidelines to manage these tumors are yet established, but complete surgical resection stands as the optimal curable treatment [8].

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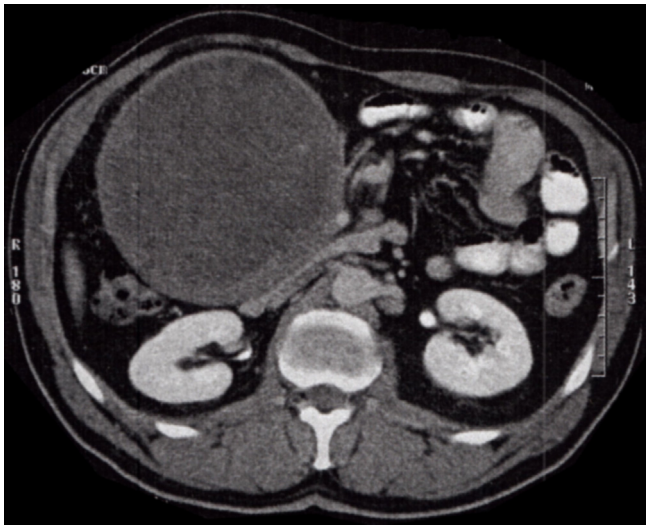

Fig. 1. Voluminous mass on the abdominal CT image.

2. Case report

A 57 years-old patient, with no relevant medical or surgical history and completely asymptomatic, was sent to our General Surgery Department after performing an abdominal contrasted computed tomography (CT) for the evaluation of a mass on the right hypochondrium. On physical examination, one could see and palpate a voluminous and indurated, yet mobile, mass.

The CT showed a single, round, 14 × 12,5 cm mass, with heterogeneous density (some small calcifications and hypodense areas), thick-walled, which compressed several intestinal loops, the uncinate process of the pancreas and also the third portion of the duodenum. It also seemed to be adherent to the third portion of the duodenum and to the gastric antrum, and a slight mesenteric densification was also present. At that point, Gastrointestinal Stromal Tumor (GIST) was the most probable diagnosis (Fig. 1).

Laboratory results were unremarkable: Hemoglobin: 15.5 g/dl, Hematocrit: 45,8%, White Blood Cell count: 7.900/mm, Platelets: 181.000/mm and normal Creatinine, Alanine Aminotransferase and Aspartate Aminotransferase levels. Tumor markers (Alfa-fetoprotein, carcinoembryonic antigen, carbohydrate antigen 19-9 and carbohydrate antigen 125) were also within the normal range of values.

Endoscopic ultrasound-guided fine needle aspiration of the mass was attempted. Nevertheless, mainly due to the notorious wall thickness, extraction of biological material for histological examination was not possible.

Taking into account that GIST was the most probable diagnosis, the patient underwent surgery, first by a laparoscopic approach and posteriorly converted to laparotomy in an attempt to completely resect the mass. A huge and well vascularized mass was found, which was adjacent to the second and third portions of the duodenum, transverse colon, body of the pancreas, and to the superior mesenteric artery and vein. Further exploration allowed to preserve these blood vessels, but a segmental colectomy of the transverse colon and a primary anastomosis needed to be performed as result of ischemia (Fig. 2). Intraoperative extemporaneous examination was performed, confirming the absence of malignancy (Figs. 3 and 4).

Definitive histology revealed a 1245 g and 14 × 14 × 10 cm mesenteric inflammatory pseudotumor, with associated ischemia, cavitary disintegration and almost complete necrosis.

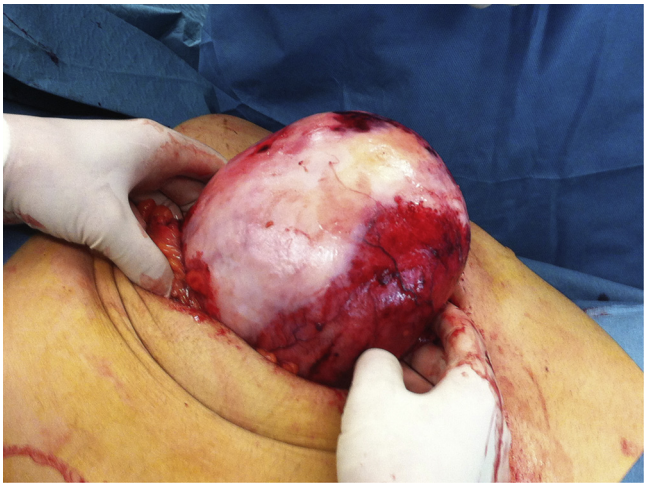

Fig. 2. Aspect of the 14 × 14 × 10 cm mesenteric IP.

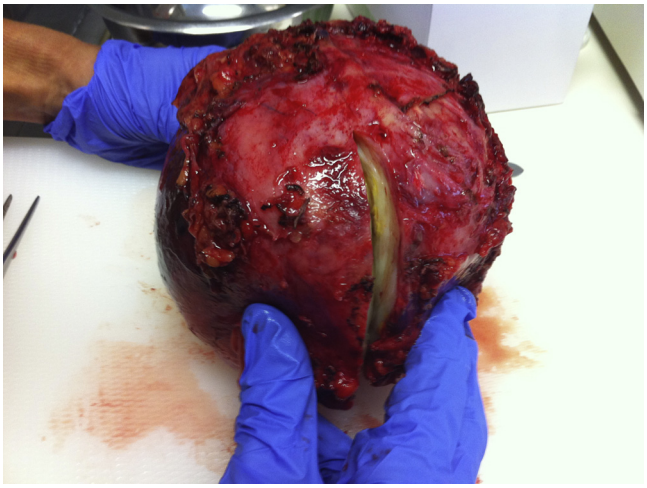

Fig. 3. Macroscopic aspect of the 1245 g IP.

The patient was discharged on the fifteenth postoperative day with no relevant morbidity. After a four-years follow-up, there is no clinical or imagiological evidence of recurrence.

3. Discussion

Preoperative diagnosis of IP can be troublesome to establish, mainly due to the lack of a typical clinical presentation.

In the literature, some retrospective reviews have been published in the pediatric population [9] whereas in adults only small series have been reported, making IP a rare entity [10]. IP can occur in nearly any organ of the human body, with the lung and the orbit being the more commonly affected ones [2,8]. Even in children, abdominal IPs are not usual and diagnosis is usually made after surgical resection [6].

Both local and systemic symptoms can be caused by the production of interleukin 1 or interleukin 6 by monocytes and macrophages [2,3]. Symptoms can be present in up to 15%-30% of the patients and are usually mild and non-specific, such as malaise, fever or weight loss. Site-related symptoms, like abdominal pain or jaundice may be present when the location of the neoplasm is within the peritoneal cavity. Usual laboratory findings are hypochromic microcytic anemia, thrombocytosis, leukocytosis, hypergammaglobulinemia and an elevated sedimentation rate [3,6].