Scholars Journal of Medical Case Reports

OPEN ACCESS

Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

Inflammatory Myofibroblastic Tumor of Digestif Truct: Three Case Reports

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DOI: 10.36347/sjmcr.2021.v09i08.012 | **Received:** 07.07.2021 | **Accepted:** 12.08.2021 | **Published:** 26.08.2021

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Abstract **Case Report**

Inflammatory myofibroblastic tumors (IMT) are a rare soft tissue and viscera neoplasm with unknown etiopathogeny. They were first described in the lung, with a wide range of synonyms. They can invade adjacent organs, recur after excision or give distant metastases. The debate persists about their inflammatory or tumor, benign or malignant nature. On the radioclinical funding, they can mimic a malignant neoplasm. The diagnosis is almost always made on the pathological examination. The treatment is poorly codified but the management is usually surgical. These tumors rarely affect the digestive truct. The only cases of gastrointestinal IMT in adults, at Hassan II university hospital in Fez during last 15 years, are those presented in this article.

Keywords: Inflammatory Myofibroblastic tumor, Digestif Truct, Adult.

INTRODUCTION

Inflammatory myofibroblastic tumor appears as a pseudotumor with a malignant manifestation. This inflammatory tumor is generally observed in children and adolescents but remains unrecognized due to its rarity. It can affect all organs in the body. The most common localization of this tumor is the lung, the mesentery comes in second position. The pathophysiology of these tumors is not yet fully understood. The manifestation of the disease is variable depending on the organ involved, and the compressive effects of the tumor are generally significant. The final diagnosis is possible through surgery and histology. Eliminating the symptoms usually requires tumor resection.

Here we will present our experience of management, within the HASSAN II University Hospital of Fez Morocco, of pseudo-inflammatory digestive tumors.

CASES REPORTS

Cas 1:

A 70-year-old patient, with no notable pathological history, who presented 7 days before admission with abdominal pain such as cramps in the right iliac fossa, associated 2 days before by an occlusive syndrome.

The patient's vital signs were: a body temperature of 39.8°C, pulse rate 112 beats/minute, respirations 24 cycles/minute, and blood pressure 110/64 mmHg. Physical examination shows distended tympanic abdomen.

A biological assessment showed correct hemoglobin at 11.3 g / dl, hyperleukocytosis at 18,300 U / mm³, a high level of C-reactive protein. The plain abdominal X-ray showed signs of small bowel obstruction.

Abdominal CT-scan showed the existence of a mesenteric mass measuring 40 * 28 mm in diameter, that comes into contact with an ileal loop which is distended and thickened with an air bubble next to it (pneumoperitoneum) (Figure 1).

The patient was admitted to the operating room, a laparatomy exploration did not show any ascites or carcinosis or hepatic metastases, and revealed an agglutination of a loop at the level of the right iliac fossa around of a small perforation of an ileal loop. The procedure consisted of an ileocecal resection and stoma

was performed. The post-operative outcome was uneventful and the patient was discharged at the fifth day.

The anatomopathological report of the specimen confirms the myofibroblastic inflammatory origin of the mass.

Cas 2

A 39-year-old man, chronic smoker, having presented 1 month before a cholestatic jaundice, associated with itching. Who presented with abdominal pain for 5 days before admission.

The patient's vital signs were: a temperature of 37.8°C, pulse 70/min, respirations 12/min, and blood pressure 138/64 mm Hg. Physical examination shows mucocutaneous jaundice accompanied by scratching lesions on the legs and abdomen.

Biological assessment revealed anemia with hemoglobin at 8.7 g/dl and perturbation of the hepatic enzymes, total bilirubin =150 mg/l, direct bilirubin= 78 mg/L, elevated levels of Aspartate transaminase = 97 U/l, Alanine transaminase = 110 U/l, and CRP = 11 mg/l.

An abdominal CT scan showed a dilation of the intra and extrahepatic duct upstream of an obstacle. (figure 2). And the abdominal MRI: showed a short stenosis of the lower bile duct suggesting a malignant process. The diagnosis of Vaterian ampulloma was retained (Figure 3).

Subsequently, the patient had undergone a duodenoscopy objectifying the presence in post-bulbar, at the level of the genu superius, of a stenosing ulcerative process. A biopsy was taken of the tumor process which was in favor of subacute nonspecific interstitial duodenitis, without seeing any tumor proliferation.

The decision of the multidisciplinary consultation meeting was surgical exploration for possible duodeno-pancreatectomy. So, the patient was admitted to the operating room, laparotomy exploration noted the absence of carcinosis or hepatic metastasis and the presence of about 3 cm tumor in the head of the pancreas with presence of a tumor of the gallbladder appearing to invade the genu superius. The procedure consisted of a Whipple-procedure accomplished with Child's anastomosis (Figure 4).

A few hours later, the patient presented a hemorrhagic shock due to a bleeding of the gastro-jejunal anastomosis requiring an urgent surgical revision. The hemostasis procedures were successful and the patient was transferred to the intensive care unit for further management. The outcome was simple and he was discharged from hospital in fourteen days.

The anatomopathological report of the specimen confirmed after immunohistochemical studies, the diagnosis of myofibroblastic inflammatory tumor.

Cas 3

18-year-old young man, with no notable pathological history, who presented 2 days before admission with abdominal pain in the right iliac fossa, not modified by eating habits or by the patient's position, without transit disorders, weight loss or associated urinary signs. Admission examination found a patient dehydrated, with tachycardia at 110 b / min and febrile at 38.5 ° C. Abdominal examination revealed a distended abdomen with palpation of a painful mass in the right iliac fossa. The patient's vital signs were a temperature of 38.8°C, pulse 100beats/minute, respirations 20cycles/min, and blood pressure at 120/64 mmHg. Physical examination shows abdomen tenderness.

A biological assessment showed hyperleukocytosis at 13 500 U / mm³, a high level of C-reactive protein. The plain abdominal X-ray showed signs of small bowel obstruction.

Abdominal CT-scan showed the existence of a large complex mass (5 x 6 cm) with heterogeneous enhancement mainly peripheral; the walls of the right colon were thickened and it was impossible to differentiate the appendix from the mass (Figure 5).

The patient had received initial antibiotic treatment with Cefotaxime, Gentamicin and Metronidazole; There had been no improvement in either the pain or the fever. Due to the aggravation of his clinic status, an emergency laparotomy was performed revealing the existence of a colonic tumor with mesocolic infiltration but without distant metastasis, a right hemi-colectomy was performed with immediate restoration of digestive continuity by an ileo-colic anastomosis (Figure 6).

The post-operative outcome was uneventful and the patient was discharged from the hospital at the fifth day. The diagnosis of an inflammatory myofibroblastic tumor was confirmed after anatomopathological study of the specimen. A two-year follow-up showed no signs of relapse.

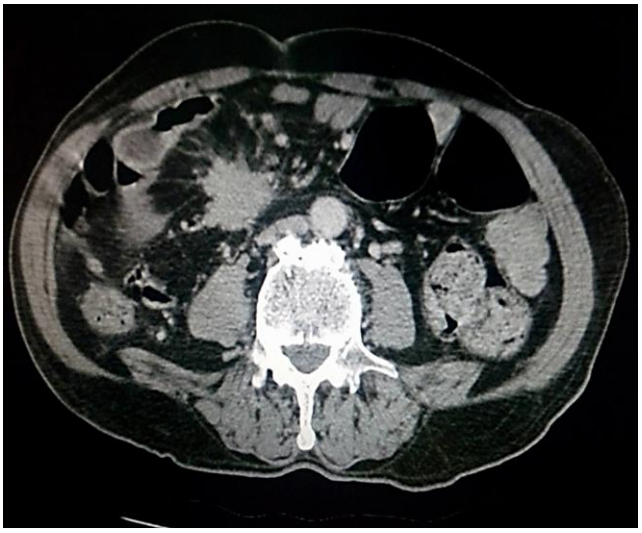

Fig-1: Abdominal CT showing the lesion at the level of the mesentery

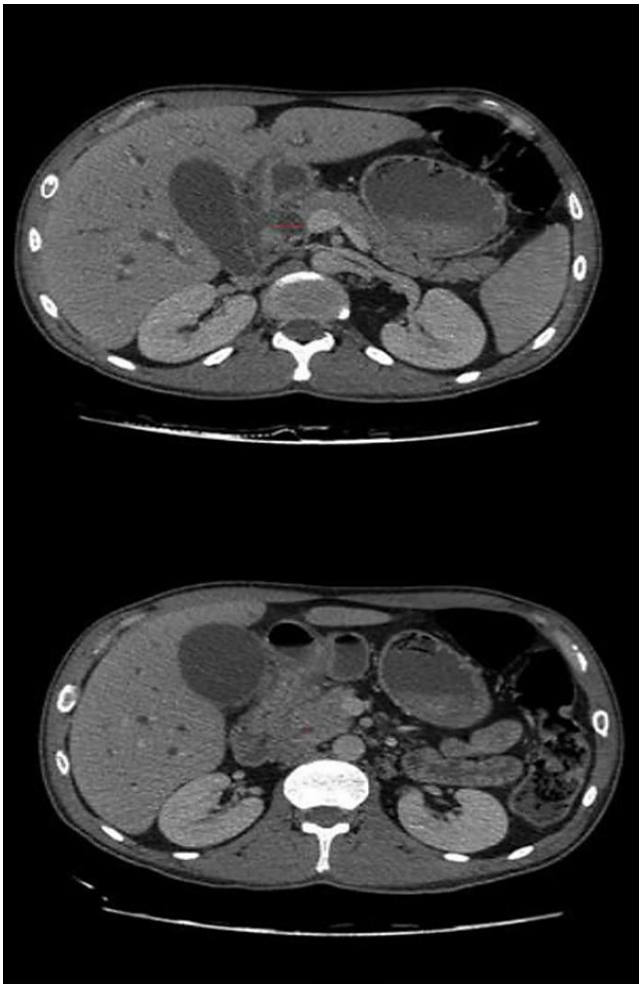

Fig-2: Axial CT scan showing the dilation of the main bile duct upstream of the tumor

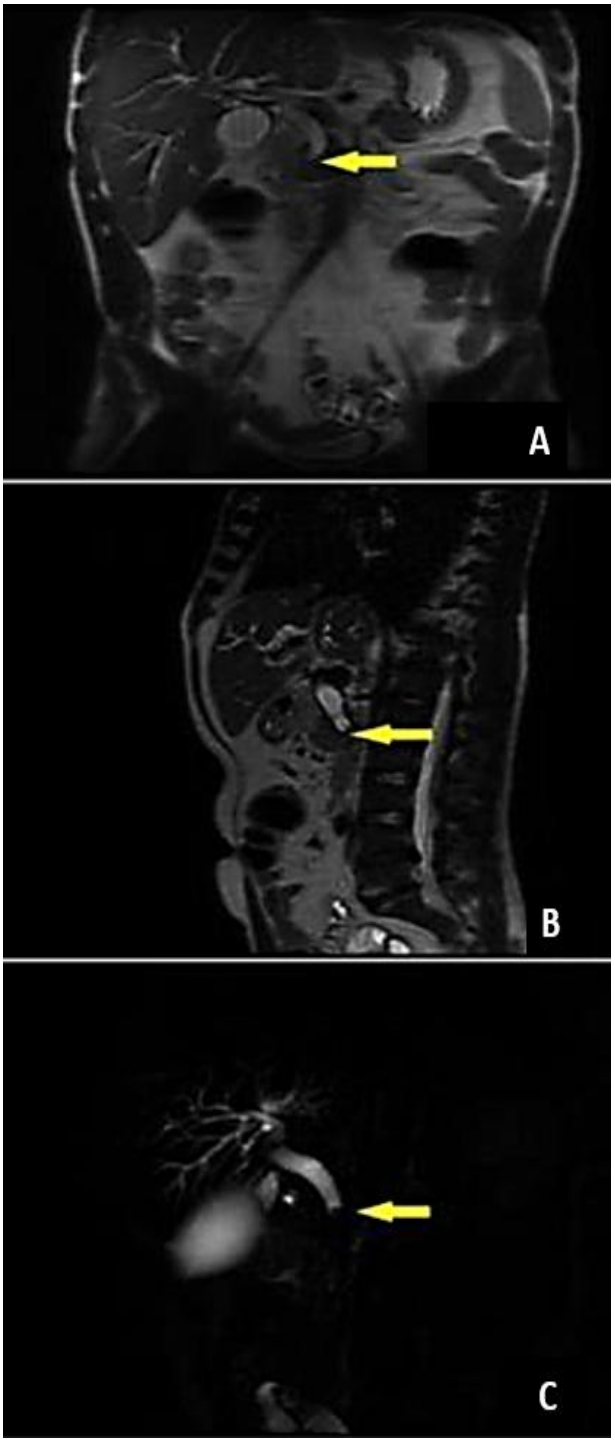

Fig-3: Abdominal MRI showing on the coronal (A) and sagittal (B) slices as well as the Bili sequence (C) a dilation of the main bile duct upstream of a short stenosis of the lower main bile duct

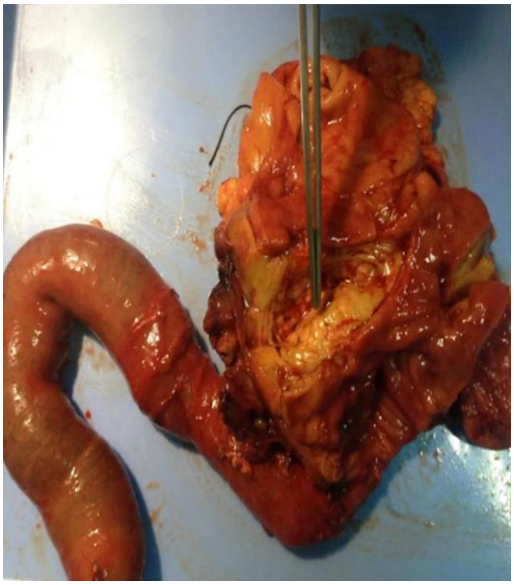

Fig-4: Image of the operative part of the cephalic duodeno-pancreatectomy showing the cephalic tumor process of the panceas

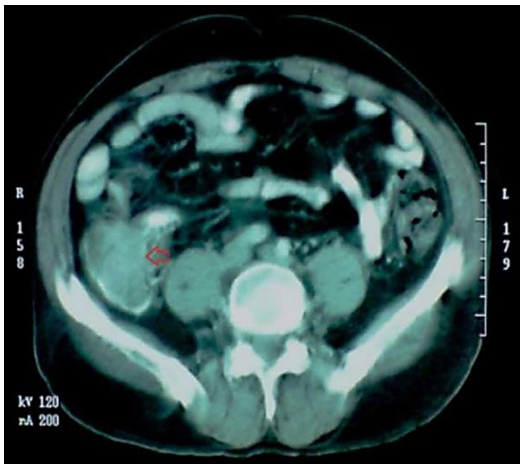

Fig-5: Abdominal CT scan showing the presence of a 6x5 cm heterogeneous colonic mass with peripheral enhancement

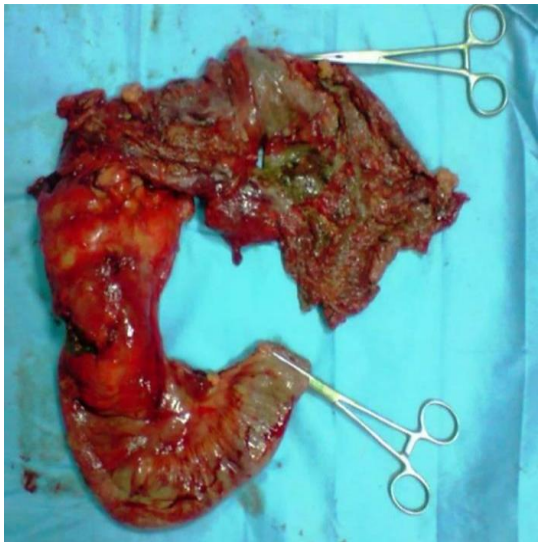

Fig-6: The operative part of the right hemi-colectomy

DISCUSSION

IMT is a rare benign tumor [1]. It is usually seen in children and adolescents aged 2 to 16 years [2]. The incidence of this tumor has been reported to be higher in girls than in boys [3]. It is possible that all organs in the body can be affected by this tumor. The most affected organs are the lung, mesentery and Omentum [4]. Given the extent of the affected area and the type of disease, this tumor is identified with different terminologies such as inflammatory pseudotumor, fibro-inflammatory tumor, eosinophilic granuloma or plasma cell granuloma [5]. However, this tumor rarely originates in the small intestine, and there are a few patients with intestinal manifestation [6]. The clinical manifestations of patients are different with respect to the affected organ, although the manifestations are rapid and progressive in bowel behaviors accompanied by symptoms such as weight loss or the emergence of a mass [7]. The cause of the disease is still unknown [8]. There are many reports on the cause of this tumor. One of these reports may be the positive EBV test [5].

It is sometimes difficult to distinguish IMT from advanced malignant tumors mainly because the mass mimics malignant tumors [2]. The initial diagnosis can be made with laboratory or radiological markers; However, surgical resection and pathological study would play an important role in the diagnosis of this type of tumor compared to other similar cases [6]. All patients who underwent surgeries were advised of long-term follow-up to investigate the risk of recurrence.

CONCLUSION

Despite the use of certain radiographic methods such as medical ultrasound tomography and computerized tomography (CT) to diagnose the disease, the definitive diagnosis is simply possible from a complete surgical resection. Final confirmation is also based on the pathological study of the specimen

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