Asian Journal of Surgery (2016) 39, 247–252

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-asianjournalsurgery.com

CASE REPORT

Inflammatory myofibroblastic tumors of the duodenum

Igor Petrovic [a], Goran Augustin [a], Ljiljana Hlupic [b], Ana Nedic [c,*], Ivan Romic [a], Mate Skegro [a]

[a] Division of Abdominal Surgery, Clinical University Hospital Centre "Zagreb", Zagreb, Croatia
[b] Department of Pathology, Clinical University Hospital Centre "Zagreb", Zagreb, Croatia
[c] Department of Internal Medicine, General Hospital "Virovitica", Virovitica, Croatia

Received 5 January 2013; received in revised form 26 July 2013; accepted 23 September 2013
Available online 6 November 2013

KEYWORDS
duodenal neoplasms;
inflammatory pseudotumor;
neoplasm;
soft tissue

Summary Inflammatory myofibroblastic tumors (IMTs) are rare soft-tissue tumors that can occur at virtually any anatomical site. We report the case of a 58-year-old male with an IMT of the fourth part of the duodenum who presented with signs and symptoms of high intestinal obstruction and bilious vomiting. The patient underwent a surgical resection of the fourth part of the duodenum with end-to-end duodenojejunal anastomosis. The follow-up period of 6 months was uneventful with no evidence of recurrence. According to our knowledge, only six cases of duodenal IMTs have been reported in the literature thus far, and this is the first report of a duodenal IMT sited at the fourth part of the duodenum. The duodenum is among the rarest sites of IMTs. Signs and symptoms resulting from diagnostic imaging investigations are nonspecific and inadequate to obtain diagnosis accurately. In most cases, surgical treatment is considered a cure for IMTs. There is no evidence of deaths caused by duodenal IMT. IMT of the duodenum is a possible diagnosis in differential diagnosis of tumor-like lesions of the duodenum.

1. Introduction

Inflammatory myofibroblastic tumors (IMTs) are rare tumors histologically composed of spindle myofibroblasts and an inflammatory infiltrate dominated by plasma cells, lymphocytes, and eosinophils.[1] According to the World Health Organization, IMTs belong to a group of soft-tissue tumors, a subset of fibroblastic/myofibroblastic tumors.[2] These principally occur in soft tissues and visceral organs at possibly any anatomical location. Nevertheless, duodenal IMTs remain an extremely rare condition. We report a case of IMT of the fourth part of the duodenum. Only six cases of

* Corresponding author. Department of Internal Medicine, General Hospital "Virovitica", Gajeva 21, Virovitica 33000, Croatia.
E-mail address: ana.nedic25@gmail.com (A. Nedic).

http://dx.doi.org/10.1016/j.asjsur.2013.09.015

duodenal IMTs have been reported in literature thus far. We present a review of these cases.[3–8]

2. Case report

A 58-year-old male who presented with a 1-week history of an intermittent epigastric pain, bilious vomiting, and unintentional weight loss of 10 kg in 4 weeks was admitted to the Department of Surgery of Clinical University Hospital Center. His medical history was significant for gastroesophageal reflux disease (GERD), *Helicobacter pylori* gastritis, Gilbert's syndrome, and acute pancreatitis of unknown exact cause 2 months ago, which was successfully treated conservatively. Multiple abdominal ultrasonographies (USs) and multislice computed tomography (MSCT) scans excluded cholelithiasis and there was no evidence of alcohol abuse in the patient's history (gamma glutamyl transferase: 17 U/L). Other causes of acute pancreatitis could neither be proven with certainty nor excluded.

A physical examination demonstrated abdominal bloating and visible distension with pain on palpation of the epigastric area of the abdomen. There was no jaundice, fever, or anemia. Laboratory tests revealed slightly elevated levels of aspartate transaminase (46 U/L), alanine transaminase (72 U/L), total bilirubin (58 μmol/L), and direct bilirubin (10 μmol/L). Other laboratory examinations analyzing the levels of C-reactive protein, erythrocyte sedimentation rate, complete blood count, coagulation factors, urea, creatinine, lipidogram, serum amylase, electrolytes including calcium, phosphorus, and magnesium were all within normal limits. Tumor markers such as cancer antigen 19-9, alpha-fetoprotein, and cancer antigen 125 were all negative.

Abdominal X-ray, abdominal US, and esophagogastroduodenoscopy showed no significant findings. Contrast imaging of the small intestine displayed a severe stricture of the ascending duodenal portion (D4) measuring 0.8 cm in diameter with normal duodenal mucosal folds and normal morphology of the horizontal and descending portions of the duodenum (Fig. 1).

The MSCT scan of the abdomen revealed a filling defect in the fourth part of the duodenum measuring 4.3 × 3.6 cm^2 and dilatation of the second and third part of the duodenum. Triangle-shaped infiltration area of the adipose tissue measuring 2.1 × 1.9 cm^2 was identified cranial to the fourth part of the duodenum (Fig. 2). No regional lymphadenopathy or focal lesions of parenchymatous organs of the abdomen suggestive of metastatic lesions were identified.

Because imaging findings could not exclude malignancy and the mass appeared to be resectable, a surgical exploration was performed through an upper midline laparotomy. A firm, elastic tumor measuring 4 cm was found in the fourth part of the duodenum, adjacent to the paraduodenal adipose tissue and peritoneum surrounding the duodenojejunal flexure. The head of the pancreas and the hepatoduodenal ligament were intact. After the right colon and the mesenteric root mobilization, superior mesenteric artery 5 cm in length was exposed, and resection of the duodenojejunal flexure measuring 15 cm in length with 2-cm surgical resection margin and clearance of loco-regional

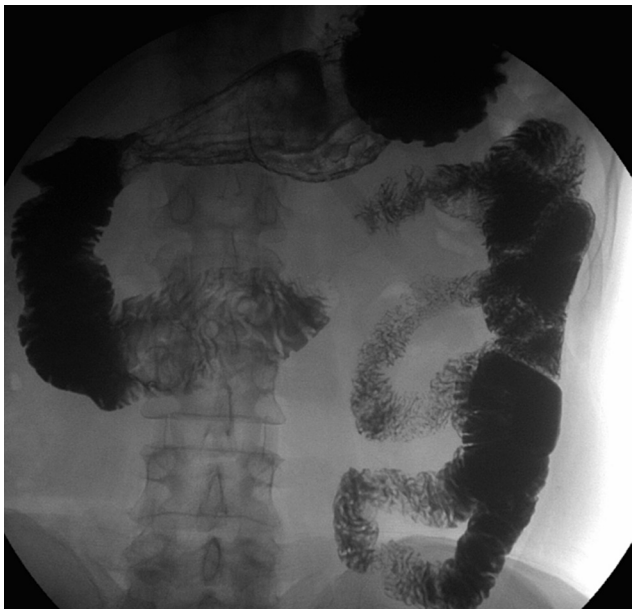

Figure 1 Contrast imaging of the small intestine displays the narrowing of the ascending duodenum.

lymph nodes were performed (Fig. 3). End-to-end duodenojejunal anastomosis with single-layer continuous suture was made for reconstruction. Macroscopic pathology demonstrated a segment of the duodenum 15.5 cm in length and 3.2 cm in diameter with exophytic polypoid tumor measuring 2.1 × 1.6 cm^2 arising from the duodenal wall (Fig. 4). Histologically, the tumor was composed of spindle-shaped interspersed myofibroblasts separated with fibrous stroma infiltrated by predominantly mononuclear inflammatory cells (Figs. 5 and 6). The tumor cells were immunohistochemically positive for actin, vimentin, AE1/AE3, and negative for nonspecific esterase, CD34, CD117, S-100, and DOG1. The resection margins were clear of malignancy. In the surrounding adipose tissue, seven lymph nodes were found, which had a diameter from 0.4 to 1.2 cm, uninvolved with tumor tissue. A postoperative

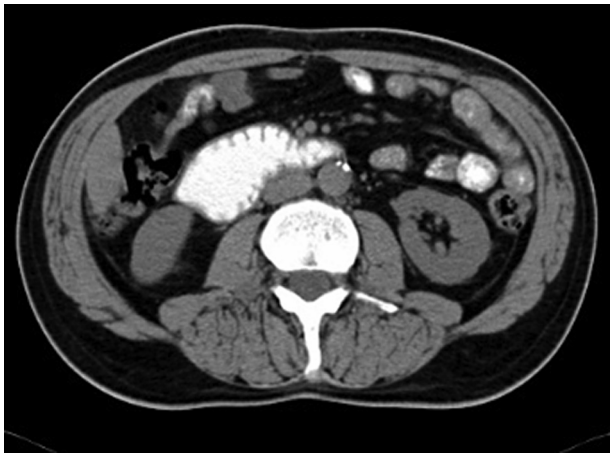

Figure 2 Multislice computed tomography demonstrates a filling defect in the fourth part of the duodenum.