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WJGS mainly publishes articles reporting research results and findings obtained in the field of gastrointestinal surgery and covering a wide range of topics including biliary tract surgical procedures, biliopancreatic diversion, colectomy, esophagectomy, esophagostomy, pancreas transplantation, and pancreatectomy, *etc*.

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CASE REPORT

Gastric inflammatory myofibroblastic tumor, a rare mesenchymal neoplasm: A case report

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Abstract

BACKGROUND

The inflammatory myofibroblastic tumor (IMT) is a rare mesenquimal tumor of doubtful biological behaviour. It's characterised for affecting mainly children and young adults, although it can appear at any age, being the lungs the primary affected organ (in children it represents 20% of all primary pulmonary tumors).

CASE SUMMARY

We present the case of a 45 year old woman, with a computed tomography (CT) finding of injury on the anterior surface of the fundus/gastric body and a solid perigastric injury of 12 mm in the ecoendoscopy. The case is presented in the tumor committee deciding to perform a laparoscopic wedge resection. The histological diagnosis was a IMT. The diagnosis is based on imaging tests like the abdominal CT, abdominal ecography and the ecoendoscopy but to confirm the diagnosis a pathological study is necessary.

CONCLUSION

Due to the unpredictable nature of this tumor, surgical resection is the best therapeutic option.

Key Words: Inflammatory myofibroblastic tumor; Gastric; Wedge resection; *ALK*-mutation; Case report

Core Tip: The inflammatory myofibroblastic tumor is a rare mesenquimal tumor of doubtful biological behaviour. It's characterised for affecting mainly children and young adults, although it can appear at any age, being the lungs the primary affected organ. The unusual thing about the case is the gastric location of the tumor (the majority are pulmonary) and the unpredictable nature of this tumor. That is why the surgical resection is the best therapeutic option.

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INTRODUCTION

The inflammatory myofibroblastic tumor (IMT) is a rare mesenquimal tumor of doubtful biological behaviour[1]. Previously known as inflammatory pseudotumor, plasmatic cell granulome, inflammatory miofibroblastoma and inflammatory miofibrohistiocityc proliferation, it's characterised for affecting mainly children and young adults, although it can appear at any age, being the lungs the primary affected organ (in children it represents 20% of all primary pulmonary tumors)[2].

CASE PRESENTATION

Chief complaints

We present the case of a 45 year old woman who requested a follow-up in our center for a second opinión after discovery of injuries that could relate to a peritoneal carcinomatosis in other center.

History of present illness

She was examined in another center given her clinic of difuse abdominal pain of long evolution without association with any other clinical manifestation, identifying in a computed tomography (CT) a "nodular isodense image, at omental level, adyacent to the anterior right abdominal wall, without discarding infiltration of the anterior right abdominal rectum. Increase in density and trabeculation of the mesenteric fat and inespecific micronodular images, findings which could relate to a peritoneal carcinomatosis".

Personal and family history

She has a history of venous insuficiency and cronic gastritis, cesarean and adenoidectomy. She denied family history of malignant tumours.

Physical examination

On physical examination, the vital signs were as follows: Body temperature, 36.8°C; blood pressure, 121/70 mmHg; heart rate, 89 beats per min; respiratory rate: 17 breaths per min. Furthermore, the patient did not have abdominal pain, but minimal ascitites in flanks was found.

Laboratory examinations

Levels of serum tumour markers were normal [carcinoembryonic antigen < 0.5 ng/mL (0.0-3.0), carbohydrate antigen (CA) 125 14.0 U/mL (0.0-35.0), CA 19-9 5.0 U/mL (0.0-40.0), CA 15-3 14.9 U/mL (0.0-28.0)]. No abnormality was found in routine blood analyses.

Imaging examinations

A gastroscopy is requested evidencing a gastric submucous injury of 1 cm in lesser curvature and a biopsy is performed. The anatomopatological diagnosis was superficial cronic gastritis not observing an intestinal metaplasia.

Subsequently, a positron emission tomography CT is performed with the following results: "Decrease in the size of the nodular image in greater omentum, with a mild affinity for FDG. Improvement of the ascitis and the trabeculation of the omental and mesenteric fat. Hepatic injury without metabolic translation".

Further diagnostic work-up

A core needle biopsy is performed of the omental injury observing fibrous tissue with no tumoral infiltration and tumor

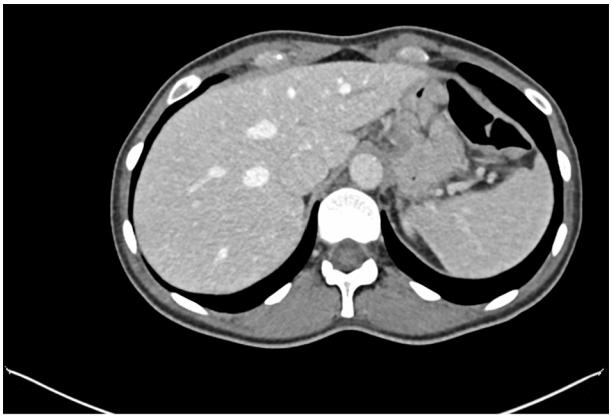

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Figure 1 Nodulary injury on the anterior surface of the fundus/gastric body.

markers are requested, which come out negative.

The control CT evidences "resolution of previous peritoneal affectation with persistence and stability of a left subdiafragmatic nodulary injury on the anterior surface of the fundus /gastric body" (Figure 1). A gastric Ecoendoscopy is requested identifying a solid perigastric injury of 12 mm with indeterminate endosonographic appearance. A biopsy is performed on 3 occasions, without obtaining a representative sample.

MULTIDISCIPLINARY EXPERT CONSULTATION

The case is presented in the tumor committee deciding to perform an exploratory laparoscopy.

FINAL DIAGNOSIS

The pathology study reported a "mesenquimal injury of 1.3 cm growing in the muscular layer of the gastric wall, which is in contact with the resection margin. It's sparsely celular, mainly constituted by fibrous tissue predominantly colagenised, with presence of spindle-shaped cells without significant atypia. Presence of psammomatous calcifications is also observed. The inmunophenotype of the tumor is: CK AE1-AE3+, Actina 1A4+ muy focal, ALK1-, Actina HHF35-, desmina-, caldesmon-, calponina-, CD34-, CKIT-, S100-" (Figure 2). Being the final diagnosis a IMT.

TREATMENT

We perform an exploratory laparoscopy. During the intervention, an injury of aproximately 2 cm is identified on the anterior gastric face, and a wedge resection of the injury is performed. The patient evolves favourably and is discharged to the second postoperative day.

OUTCOME AND FOLLOW-UP

At 1 mo postoperatively, the patient was still alive. Given these findings, the case is presented again to the multidisciplinar committee, which decides to carry out an annual Ecoendoscopy and toracic CT to examine the pulmonar nodule.

DISCUSSION

The IMT is rare tumor, predominantly located in the lungs but can also be found in the retroperitoneum, mesentery, head, neck and stomach. The latter case, presented by our patient, is extremely rare, with very few described in literature (Table 1).

It is characterized by local recurrence but rarely incurs in distant metastasis[3]. Risk factors for the development of IMT have not been established, but cases have been described which suggest association with Virus Epstein bar, genetic alterations like the reorganisation of the anaplastic lymphoma kinase (*ALK)* gene in the 2p23 cromosome or alterations of the inmune system[3,4]. They are usually asymptomatic or present inespecific symptoms like abdominal pain, toracic

Table 1 Clinicopathological characteristics of inflammatory myofibroblastic tumors in adults, described in the literature

Ref.	Sex/age	Presenting symptoms	Tumor localization in the stomach	Tumor size (cm)	Mitosis	Histologic pattern	Treatment	Follow-up
París-Sans *et al*[1]	M/88	AP, vomiting, jaundice	GDJ	4	/	Proliferation of spindle-shaped mesenchymal cells mixed with lymphocytes	PG	/
Cheng *et al* [3]	W/52	AP	Antrum (exophytic)	4.3	0-1	Proliferation of fusiform cells	DG	6 mo
Bjelovic *et al* [4]	W/43	AP, nausea	Distal Stomach	6	44928	Hypercellular spindle cell proliferation with vague fascicular areas	DG	2 yr
Shi *et al*[5]	M/36	AP, AM	Antrum, LC	4.5	44928	Myxoid hypocellular with some fascicular areas	PG	5 yr (NED)
Shi *et al*[5]	M/42	AP, UGH, AM,	Upper body, GC	8	44928	Fascicular with some myxoid areas	PG	Recurrence at 12 mo
Shi *et al*[5]	M/40	AM	Upper body, AW	6.3	44928	Myxoid hypocellular with some fascicular areas	PG	3 yr (NED)
Shi *et al*[5]	M/45	AP, AM	Angle	5.5	44928	Myxoid hypocellular with some fascicular areas	PG	2.6 yr (NED)
Shi *et al*[5]	W/45	AP, AM	Lower body, PW	5.8	44928	Fascicular with some myxoid and sclerotic areas	PG	4 yr (NED)
Katakwar *et al*[6]	M/45	AP	AW	5	44928	Hypocellular, collagenized, myofibroblatic cells	DG	Recurrence at 1 mo
Leon *et al*[7]	W/50	Vomiting, weight loss	PW	7	44928	Patternless round and spindle cell proliferation	PG	2 yr (NED)
Park *et al*[8]	W/55	AP, hematoperitoneum	Upper body, GC	8	44928	Vague fascicular proliferation	Gastric wedge resection	/
Jadhav *et al* [9]	M/18	AM, weight loss	LC	9	44928	Pleomorphic cells, spindle-shaped to stellate cells arranged in a background of myxoid	Excision	5 yr
Qiu *et al*[10]	W/61	Fever	LC	3	/	Spindle cells with inflammatory infiltrate of neutrophils, eosinophils, lymphocytes, and plasma cells.	DG	3 mo (NED)
Kim *et al* [11]	M/25	AM	GEJ	8	/	/	/	/
Albayrak *et al*[12]	W/56	Nausea, vomiting, UGH	Cardia	11	44928	Granulation-type and storiform spindle cell proliferation	PG	8 mo (NED)
Our Study	W/ 45	AP	AW (exophytic)	1.3	/	Spindle/stellate cells with inflammatory cells	Gastric wedge resection	1 mo

AP: Abdominal pain; AM: Abdominal mass; UGH: Upper gastrointestinal hemorrhage; LC: Lesser curvature of the stomach; GC: Greater curvature of the stomach; AW: Anterior wall of the stomach; PW: Posterior wall of the stomach; C: Cardia; PG: Partial gastrectomy; DG: Distal gastrectomy; NED: No evidence of disease; GDJ: Gastroduodenal junction; GEJ: Gastroesophagus junction; M: Male; F: Female.

pain, and up to 30% develop a constitutional syndrome[1,2].

The diagnosis is based on imaging tests like the abdominal TC, abdominal ecography and the ecoendoscopy[5].

The differential diagnosis includes gastrointestinal stroma tumor, fibroid inflammatory polyp, single fibrous tumor or peripheral nerve tumors, amongst others[6].

Confirmation diagnosis is obtained with histological examination, which evidences proliferations of myofibroblasts, lymphoplasmacytic infiltrate, and a myxoid stroma[5].

Fifty-six percent of IMT present reorganisation of gene ALK. These patients present a higher risk of local recurrence but not distant metastasis (negative ALK), which suggests that reactivity to the ALK could be a protective factor[3].

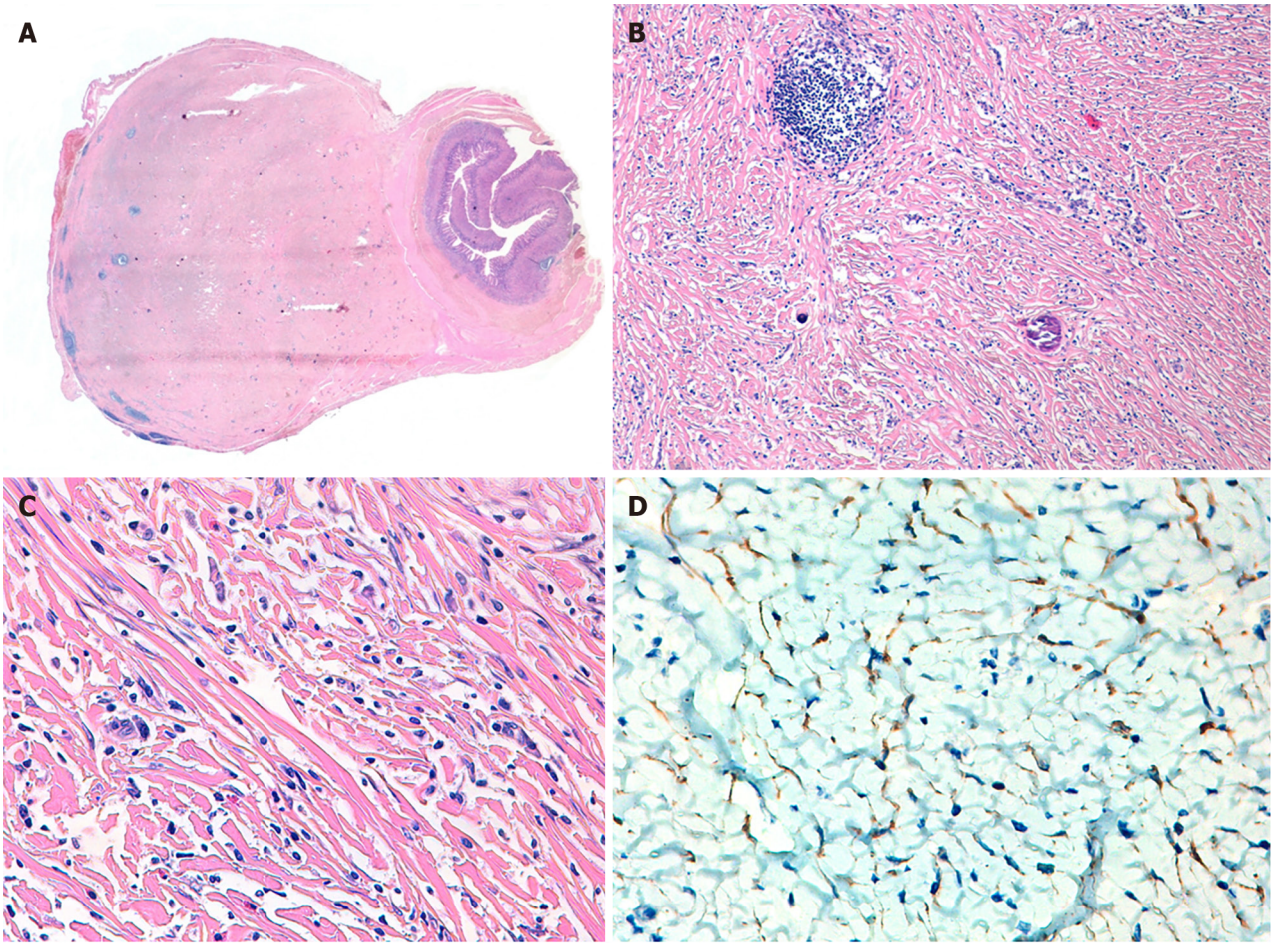

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Figure 2 Histopathological analysis and immunohistochemical examination of the resected specimen. A: Panoramic image of the lesion; B: Mesenquimal injury and psamomatous calcifications; C: Mesenquimal lesion with infiltrate of eosinophils, plasma cells and masts cells; D: Positive expression of Cytokeratins AE1-AE3.

CONCLUSION

Due to the unpredictable nature of this tumor, surgical resection is the best therapeutic option. Regarding gastric IMTs, depending on the tumor's location, options go from a wedge resection to a partial gastrectomy[1-3]. Patients which cannot undergo surgical interventions, can be treated with a combination of radiotherapy and chemotherapy. Patients with metastatic tumors or local advanced tumors resistent to conventional chemotherapy can be treated with Crizotinib if they present a mutation of ALK or Larotrectinib o Entrectinib, if they present mutations in the gene TRK[2].

The recurrence rate in the first year after surgery is of 15%-37%, therefore clinical and radiological follow-ups are indicated, without finding in the literature a defined periodicity for them[3,4].

FOOTNOTES

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