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CASE REPORT

Remission of Feline Gastrointestinal Eosinophilic Sclerosing Fibroplasia in a Cat Treated with Corticotherapy

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ABSTRACT

Feline Gastrointestinal Eosinophilic Sclerosing Fibroplasia (FGESF) is a rare clinical entity of unknown aetiology. The most defended hypothesis refers to a genetic alteration in the immune response regulation, which results in an exacerbated eosinophilic inflammation. The proposed treatment for FGESF includes immunosuppressive drugs and surgical resection of the lesion. A 2- and- a- half-year old neutered, male, Chartreux cat was diagnosed with FGESF with the presence of a mass in the first duodenal flexure, which was surgically removed and recurred 8 months post-surgery. After using an immunosuppressive treatment for one year, the macroscopic disappearance of the lesion and the complete remission of the clinical signs were achieved. To our knowledge, this is the first description of disappearance of a FGESF lesion located in the gastrointestinal tract after prolonged immunosuppressive treatment. This clinical report highlights the possibility to treat this new feline disease with the exclusive use of immunosuppressive drugs.

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INTRODUCTION

Feline Gastrointestinal Eosinophilic Sclerosing Fibroplasia (FGESF) was described in 2009 as a rare entity that could be included into the inflammatory eosinophilic syndrome (Craig *et al.,* 2009). The disease is characterized by the presence of masses limited to the gastrointestinal tract and associated lymph nodes in most affected cats with specific histological features (Craig *et al.,* 2009, Linton *et al.,* 2015). These lesions have been recently described in other locations: pancreas, liver, mesentery or abdominal cavity (Weissman *et al.,* 2013, Thieme *et al.,* 2019, Kambe *et al.,* 2020).

The aetiology of FGESF remains unknown, although a genetic alteration in the regulation of the immune response resulting in an exacerbated eosinophilic inflammation has been proposed (Craig *et al.,* 2009, Weissman *et al.,* 2013, Linton *et al.,* 2015). This eosinophilic response translates into a fibroblastic proliferation with an extracellular matrix deposit, forming over time the characteristic masses of this disease (Gomes *et al.,* 2005). The finding of intralesional bacteria in a high percentage of cases, as well as other organisms, such as fungus or parasites (Ozaki *et al.,* 2003; Grau-Roma *et al.,* 2014), suggests some role for these agents in the pathogenesis of the disease (Craig *et al.,* 2009; Linton *et al.,* 2015).

The proposed treatment for FGESF usually includes a multimodal approach consisting of immunosuppressive therapies, according to the suspected immune-mediated etiology, following the surgical resection of masses (Craig *et al.,* 2009; Linton *et al.,* 2015).

This case report describes a cat diagnosed with FGESF with the presence of an intestinal mass, which was surgically removed and recurred 8 months post-surgery. After using an immunosuppressive treatment for one year, the macroscopic disappearance of the lesion and the complete remission of clinical signs was achieved. The possibility to manage the gastrointestinal presentation of this disease exclusively with medical treatment without a surgical intervention is discussed.

Case history: A 2- and- a- half- year old neutered, male, Chartreux cat was presented with a 6-month history of

twice a day partially digested food and hair remnants vomiting few hours after the intake. Physical examination did not reveal remarkable findings, abdominal palpation was normal and body condition score was 7/9. The cat was negative for feline leukemia virus and feline immunodeficiency virus, did not show anorexia or weight loss, neither any other clinical sign or alterations in complete blood count (CBC), serum biochemical profile and urinalysis.

The serial coprology analysis was negative and abdominal radiography and ultrasound did not show any alteration. After one month with a hydrolyzed protein diet (Royal Canin Hypoallergenic, Missouri, EEUU) and daily brushing, the cat went on vomiting with a lost weight of 10%. During upper digestive endoscopy, it was observed a 10x10 mm intramural ulcerated mass located in the duodenum, 0.5 centimeters from the pyloric sphincter (Fig. 1), producing a partial intestinal lumen obstruction. Endoscopy biopsies were collected from the lesion, other duodenal areas and the stomach. The mass displayed a lost structure in the deep lamina propria due to the presence of dense connective tissue areas with fibroblastic proliferation, which displaced the glandular tissue. Associated with these fibrotic masses, a moderate to severe inflammatory process was observed, with numerous eosinophils, and plasma cells, lymphocytes and mast cells in a lower proportion. Similar inflammatory response was observed in the lamina propria of fundus, gastric body, and duodenum. The accurate diagnosis was FGESF and an exploratory laparotomy was conducted.

At exploratory laparotomy a firm and focal nodule was detected in the proximal duodenum close to the pancreatic duct. Due to the location of the mass and considering the instauration of medical therapy after the surgery, a conservative surgical mass resection was chosen to minimize the risk of surgery-related sequelae. Complete inspection of the abdominal cavity did not reveal other lesions including lymphadenomegaly. Histopathology study of the samples stained with hematoxylin and eosin, Masson´s trichrome and toluidine blue techniques confirmed FGESF. The lesion exhibited the characteristic trabecular pattern of dense collagen separated by fibroblasts and infiltrate with a mixed population of inflammatory cells with predominance of eosinophils and mast cells, and fewer lymphocytes, plasma cells and neutrophils (Fig. 2). Neither bacterial nor fungal agents were detected. Culture techniques were also employed, using selected media for aerobic, anaerobic (blood agar medium) and gram-negative (MacConkey medium) bacteria and for fungal growth (Sabouraud agar medium), but no organism grew.

The post-surgical antibiotic treatment was metronidazole (10mg/ kg PO every 12 hours for 15 days) and cephalexin (22mg/ kg PO every 8 hours for 15 days). Fifteen days after the surgery, prednisone (1mg/kg PO every 12 hours in decreasing doses during 90 days) was added to the pre-established dietary treatment. The cat remained asymptomatic for eight months. Nevertheless, at the end of this period, daily vomiting reappeared. A new ultrasound study revealed mesenteric lymphadenopathy of 1.3-1.5 cm and a 6 mm thickening in the muscular layer of the stomach with increased echogenicity and normal wall layering. The macroscopic stomach appearance during endoscopy was completely normal. In contrast, the recurrence of a mass with the same characteristics, diameter and location was evidenced in the duodenum. The histological study of the biopsies confirmed the diagnosis of FGESF.

On this occasion, a conservative medical treatment was chosen due to the decision of the owner. Together with the hydrolyzed protein diet, the immunosuppressive treatment with prednisone was restarted at minimum effective dose (0.2 mg/kg PO every 12 hours) for one year. After completion of the treatment the cat remained asymptomatic, and an upper digestive endoscopy was performed, evidencing the regression of the lesion (Fig. 3). Histologically, the study of biopsies taken in the area where the lesion was located determined the presence of an inflammatory lymphoplasmacytic infiltrate with marked fibrosis. Seventeen months after the last endoscopy, the cat continues in clinical remission with hydrolyzed protein diet and prednisone at minimum effective dose at the time of writing.

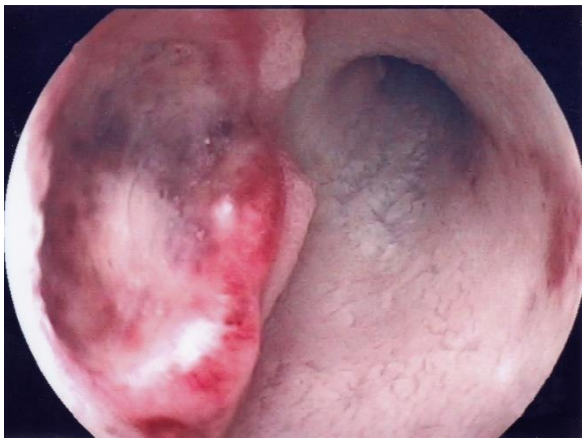

Fig. 1: Endoscopic image of a ten millimeters in diameter, intramural and ulcerated mass located in the duodenum, 0.5 centimeters from the pyloric sphincter that produced a partial intestinal lumen obstruction.

DISCUSSION

The typical presentation of FGESF consists of gastrointestinal masses characterized by trabecular pattern of dense collagen, fibroblasts and a mixed inflammatory infiltrated with a predominance of eosinophils and abundance of mast cells (Craig *et al.,* 2009). There is no breed, sex or age predisposition (Craig *et al.,* 2009; Weissman *et al.,* 2013; Linton *et al.,* 2015). Our case is the first report of FGESF in a Chartreux cat.

As all the cases reported, the cat showed nonspecific digestive signs, that could be caused by the inflammation of the gastrointestinal tract, the semi-obstructive effect of the mass or both (Craig *et al.,* 2009; Jergens, 2012; Linton *et al.,* 2015). As in this case, the absence of the most reported FGESF´s findings (palpable abdominal mass, eosinophilia and hyperproteinaemia) could not allow ruling out the disease (Craig *et al.,* 2009; Weissman *et al.,* 2013; Linton *et al.,* 2015).

Imaging techniques such as radiography or abdominal ultrasound are not always able to show these lesions and digestive endoscopy or computerized tomography are needed (Weissman *et al.,* 2013; Thieme *et al.,* 2019; Kambe *et al.,* 2020).

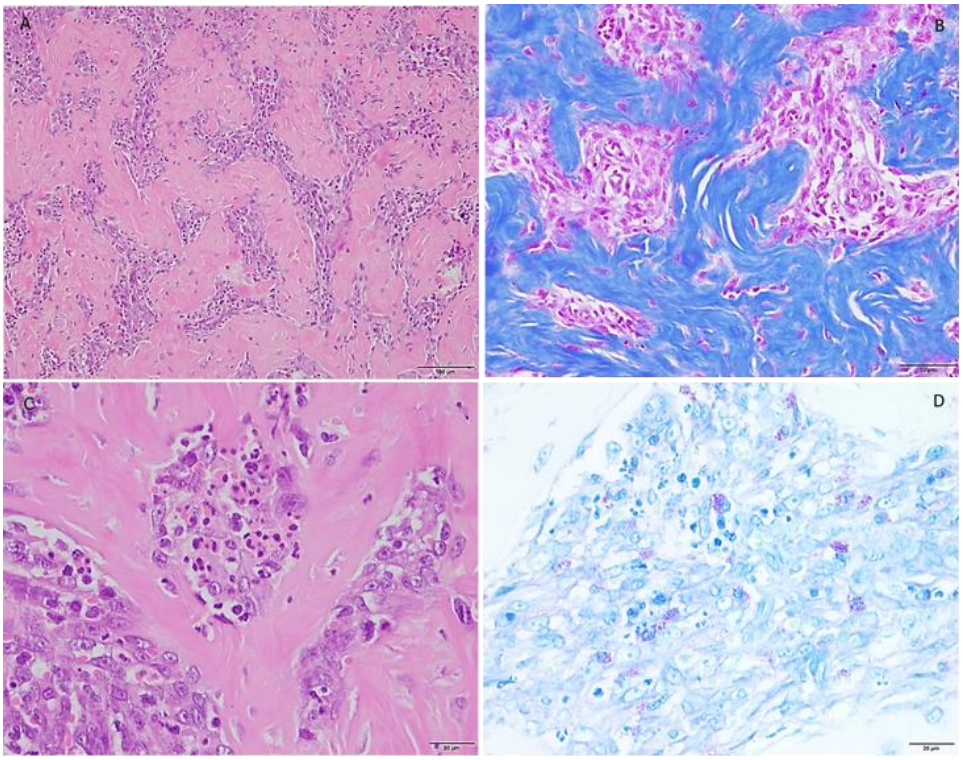

Fig. 2: A: Histological appearance of the duodenal mass characterized by dense branched and anastomosed collagen trabeculae separated by an abundant population of fibroblasts and a mix inflammatory infiltrate. H&E (magnification: x10), B: With high magnification dense blue connective bands can be seen separated by small inflammatory foci. Masson´s trichrome stain (magnification: x20). C: Inflammatory foci were characterized by the majority presence of numerous fibroblast and overall eosinophils together with lymphocytes and plasma cells. H&E (magnification: x40). D: Duodenal mass displayed in the proliferative fibroblastic areas the presence of a high number of dispersed and perivascular mast cell. Toluidine blue technique (magnification: x40).

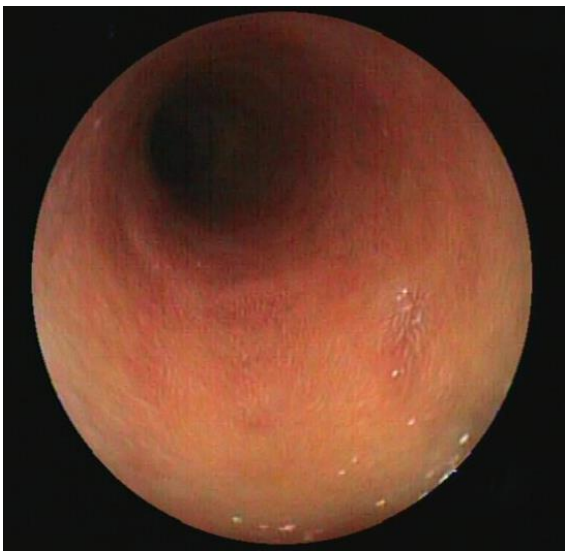

Fig. 3: Endoscopic image of the duodenum where the macroscopic regression of the lesion located there one year before can be evidenced. Duodenal mucosa appears slightly thickened and congestive.

Although a possible genetic etiology of the disease could not be discarded, neither the parents nor the siblings of the cat showed any digestive clinical sign.

Bacteria were not detected in this case. Under normal conditions no bacteria would be found in a gastrointestinal intramural lesion, and only 3-5% of the bacteria from the intestinal microbiota grow in culture (Suchodolski, 2011). Due to this and the disagreement amongst the microbiological detection techniques used in the published cases (Linton *et al.*, 2015), future research with mass sequencing techniques are needed in order to clarify the role of microorganisms in the disease.

Variable prognosis is considered for affected cats and it depends on the location of the masses and the possibility of surgical resections, an early diagnosis and the treatment administered (Craig *et al.*, 2009; Weissman *et al.*, 2013; Linton *et al.*, 2015). Even considering that the surgical approach is the first step usually recommended, it is not always easy to implement based on the literature findings. Intraoperative and perioperative mortality is remarkable in the surgical procedure of FGESF (Craig *et al.*, 2009; Linton *et al.*, 2015). In a case series of 25 cats, 24% of patients were euthanized during the laparotomy (Craig *et al.*, 2009). Sometimes masses cannot be resected due to their size or their location, especially when the mass is found near the pyloric region and the duodenal papilla. In those cases where surgery is not possible or a complete resection of the mass is not achieved, Linton and co-workers suggest the subsequent use of immuno-suppressive therapy in order to increase the survival rate (Linton *et al.*, 2015). Taking into account the risks of the surgery in many cases of FGESF and the favourable evolution obtained exclusively with medical therapy in the relapse of the case presented in this study, a prolonged immunosuppressive treatment could be recommended as a first-choice therapy in cases suffering from this disease, as occurs in different entities included into the inflammatory eosinophilic syndrome. Similar remission of clinical signs has been recently described after medical therapy in two cats with FGESF with masses located outside the gastrointestinal tract (Thieme *et al.*, 2019; Kambe *et al.*, 2020), supporting the potential utility of this therapeutic strategy also in intraluminal digestive masses.

Conclusions: This clinical report highlights the importance of including this new feline disease in the differential diagnosis of nonspecific gastrointestinal signs in concurrence with gastrointestinal mass. A prolonged immunosuppressive treatment resulted in this case in the disappearance of the lesion and the clinical signs, suggesting the possibility to treat this entity only with immunosuppressive therapies decreasing the mortality associated to a surgical resection of the lesion placed

complicatedly. Furthermore, a longer follow-up and more cases are required to better understand FGESF.

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Authors contribution: BA and DDR collected the cat´s follow-up and wrote the manuscript. AV wrote the manuscript. MGS, FRF and AS carried out the diagnosis, instituted the treatment and carried out the clinical follow-up. CPD performed the surgery. ARB carried out the histopathological diagnosis. All authors critically revised the manuscript for important intellectual contents and approved the final version.

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