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Feline gastrointestinal eosinophilic sclerosing fibroplasia presenting as a rectal mass

Laura M Goffart[1], Alexane Durand[2], Martina Dettwiler[3] and Simona Vincenti[1]

Abstract

Case summary A 9-year-old neutered male cat was referred owing to dyschezia and weight loss. Abdominal CT revealed a heterogeneous mass in the rectum and thickening of one caudal mesenteric lymph node. The mass induced a focal rectal obstruction. Cytological evaluation of fine-needle aspirates showed signs of mixed inflammation for the rectal mass and a reactive lymph node. Because a definite diagnosis was not achieved, complete resection of the mass via a dorsal approach to the rectum was attempted. Histopathology confirmed complete removal and diagnosed feline gastrointestinal eosinophilic sclerosing fibroplasia (FGESF). The cat was treated with psyllium husks and lactulose after surgery. In the postoperative year, the owner reported normal behaviour, food intake and defecation of the patient. Dyschezia reoccurred 14 months after surgery. Imaging revealed recurrence of a rectal mass. Owing to clinical deterioration, the owner elected for euthanasia.

Relevance and novel information This is the first report of rectal FGESF with dyschezia and weight loss as the main clinical signs. The case demonstrates an acceptable outcome for more than 1 year without additional immunosuppressive therapy, and emphasises that FGESF must be considered as a differential diagnosis for rectal masses in cats.

Keywords: FGESF; rectal masses; rectal surgery; oncology

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Case description

A 9-year-old male neutered domestic shorthair cat was referred to the Small Animal Clinic of the Vetsuisse Faculty, University Bern, for dyschezia, weight loss and hyporexia. The primary care veterinarian had treated the cat for 3 days with meloxicam (0.05 mg/kg PO q24h), antibiotics (amoxicillin 20 mg/kg PO q12h), subcutaneous injections of saline and enemas. Owing to a lack of improvement in the dyschezia, and the detection of a caudal rectal mass upon rectal palpation, the cat was referred for further treatment. At the time of referral, the cat had shown dyschezia for 1 week and the last normal defecation had been noted 3 days previously.

On the day of presentation, the cat was bright and alert. Physical examination was within normal limits. The cat had a body weight of 4.6 kg and a body condition score of 6/9. Abdominal palpation was unremarkable. Despite being in a good nutritional status, the owner reported that the cat had lost roughly 500 g of body weight over the past month.

Full haematology results were unremarkable. Serum chemistry showed mild hyperglycaemia (7.8 mmol/l; reference interval [RI] 3.17–5.71), marginal hyperglobulinaemia (42.1 g/l; RI 26–42), mildly elevated aspartate

[1]Division of Clinical Surgery, Vetsuisse Faculty, University of Bern, Bern, Switzerland
[2]Division of Clinical Radiology, Vetsuisse Faculty, University of Bern, Bern, Switzerland
[3]Division of Clinical Pathology, Vetsuisse Faculty, University of Bern, Bern, Switzerland

Corresponding author:
Simona Vincenti DVM, Division of Clinical Surgery, Vetsuisse Faculty, University of Bern, Länggassstrasse 128, Bern 3012, Switzerland.
Email: simona.vincenti@vetsuisse.unibe.ch

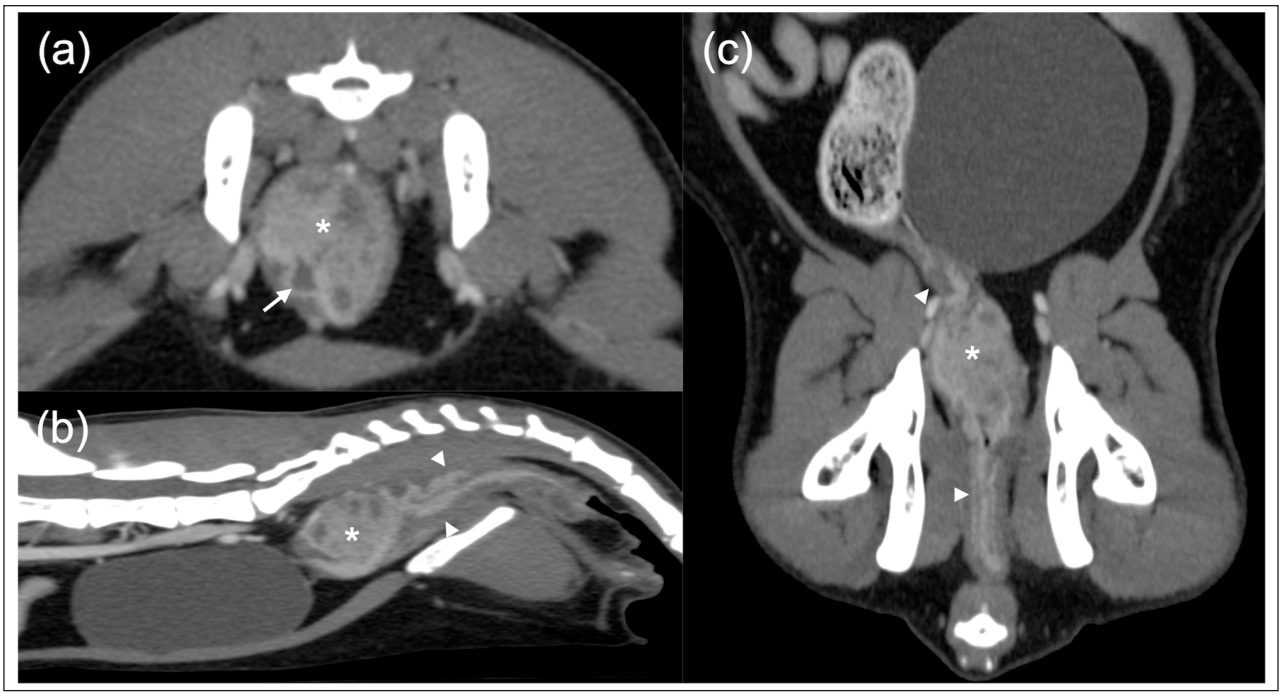

Figure 1 Post-contrast (a) transverse, (b) sagittal and (c) dorsal CT images of the pelvic canal of an adult cat, showing a large, asymmetrical, eccentric, heterogeneously enhancing, soft tissue-attenuating transmural rectal mass (asterisk), leading to severe luminal narrowing (arrow). Mild circumferential thickening of the surrounding distal descending colon and rectal wall was noted (arrowheads)

aminotransferase (97 U/l; RI 12–61), markedly elevated creatine kinase (5710 U/l; RI 0–596) and slightly decreased urea (5.6 mmol/l; RI 6.5–12.2).

To perform a thorough rectal examination and CT (Philips Brilliance CT 16-slice scanner; Philips AG Healthcare), the cat was premedicated with butorphanol (0.2 mg/kg IV) and medetomidine (0.005 mg/kg IV), and general anaesthesia was induced with propofol (2 mg/kg IV). Dual-phase abdominal CT revealed a large, eccentric, asymmetrical, heterogeneously enhancing, soft tissue-attenuating transmural rectal mass of approximately $3.4 \times 2 \times 2.2$ cm in size (Figure 1). The mass generated a marked luminal narrowing with focal loss of visualisation of the rectal lumen. A mild homogeneous circumferential thickening of the surrounding distal descending colon and rectal wall was noted. Additionally, one of the caudal mesenteric lymph nodes was moderately thickened (6.5 mm) and elongated, and a minimal amount of free fluid was visible in the caudal abdomen. Considered differential diagnoses included a primary rectal neoplasia (eg, lymphoma and carcinoma) or a rectal inflammatory granuloma. The regional lymphadenopathy was either reactive or of metastatic origin.

For further evaluation, ultrasound-guided fine-needle aspiration of the mass and enlarged lymph node was carried out. The rectal mass cytology revealed a mixed inflammation, including a high number of degenerated neutrophils containing intracellular rod-shaped bacteria, low numbers of eosinophils, individual well-granulated mast cells, and probable reactive fibroplasia. The lymph node was reactive, showing no signs of malignancy or microorganisms.

After a thorough discussion with the owner, surgical resection of the mass with wide margins was elected. The cat was premedicated with dexmedetomidine (0.005 mg/kg IV) and methadone (0.22 mg/kg IV). General anaesthesia was induced with ketamine (1 mg/kg IV) and alfaxalone (0.65 mg/kg IV) and maintained with isoflurane in air oxygen mixture. Ampicillin sulbactam (30 mg/kg IV) was administered preoperatively and then repeated every 90 mins perioperatively. Additionally, the cat received an epidural anaesthesia (ropivacaine 0.98 mg/kg combined with morphine 0.1 mg/kg and methadone 0.1 mg/kg). The patient was positioned in ventral recumbency with the tail pulled and fixed over the back, the pelvis elevated and the hindlimbs padded against the table. After surgical scrub, the perineal area was draped.

A dorsal approach to the rectum was performed. A crescent-like skin incision was performed dorsally to the anus. Then, the rectococcygeal muscles were transected after placement of a holding suture with Prolene 4-0 (Prolene 4-0; Ethicon) on the dorsal aspect of the muscle, to ease the final muscles' reconstruction. The caudal and middle rectum were carefully isolated from the surrounding tissue using digital and blunt dissection. Afterwards, three holding sutures (Prolene 4-0) were

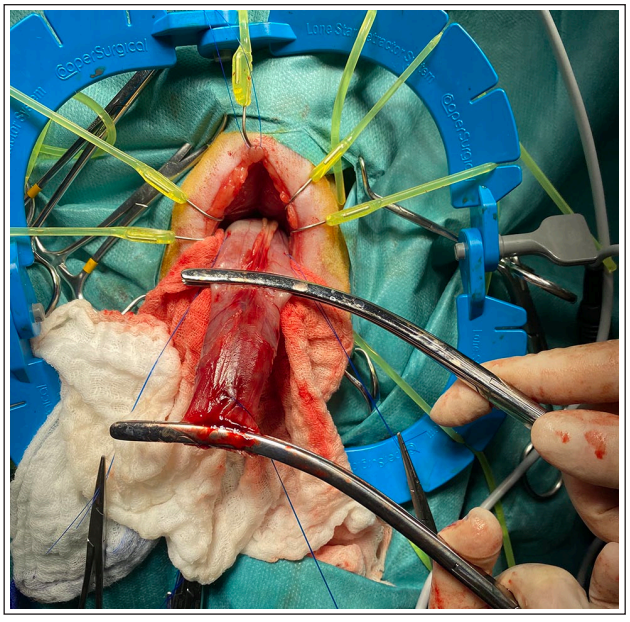

Figure 2 Intraoperative view 1. Positioning of the Doyen clamps orally and aborally to the mass leaving 1 cm of palpable margin to the mass

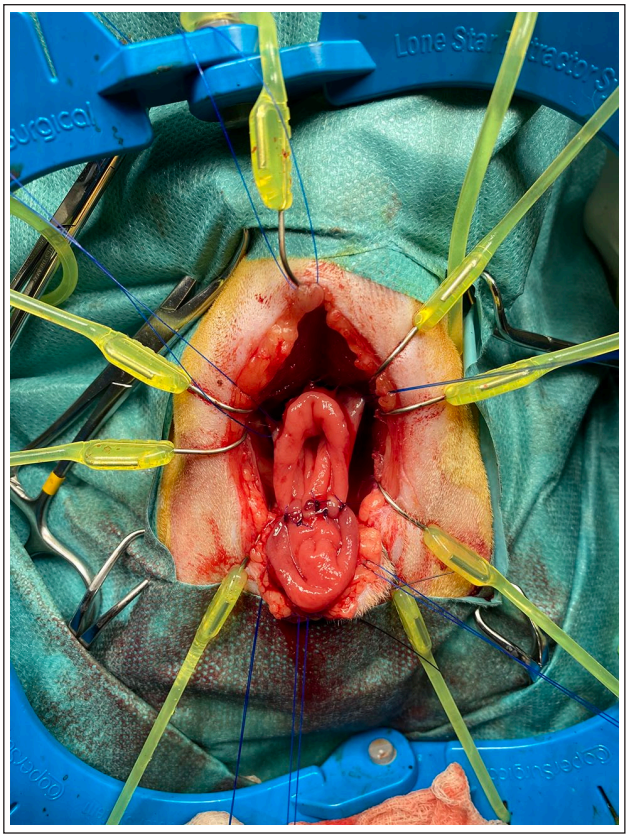

Figure 3 Intraoperative view 2. Generation of the anastomosis beginning ventrally. The knots of the ventral sutures lie intraluminally

placed at both the caudal and the cranial edges of the intended enterectomy site, which was elevated dorsally using Ethiloop (Ethiloop 2 mm; Ethicon). Two Doyen clamps were placed next to the intended enterectomy margins (Figure 2), and sharp transection of the rectum was performed taking at least 1 cm margin on both sides of the rectal mass. After completion of the enterectomy, gloves and instruments were changed, and the surgical field was profusely flushed with lukewarm sterile saline. Starting ventrally, a single-layer, simple-interrupted rectal anastomosis was performed using PDS 4-0 (PDSII; Ethicon) (Figure 3). Intraluminal knots were placed at the ventral aspect, while extraluminal knots were placed at the dorsal and both lateral aspects of the rectal anastomosis. After completion of the anastomosis (Figure 4), and copious lavage of the surgical area, the rectococcygeal muscles were re-apposed using simple interrupted suture, and subcutaneous and cutaneous tissues were routinely closed. The cat was then placed in right lateral recumbency to place a left-sided oesophageal feeding tube. The patient recovered uneventfully from anaesthesia.

Postoperative therapy included buprenorphine (0.02 mg/kg q8h IV) for 4 days, psyllium husks (2 g/kg q12h) and lactulose (1 ml PO q12h). Parenteral nutrition (Royal Canin Recovery Liquid Dog/Cat) with 200 kcal daily was administered through the oesophageal feeding tube for the first 6 days after surgery.

One week postoperatively, the cat was discharged from the hospital showing normal vital parameters, no signs of infection or stricture at the surgical site, normal urination and regular defecation with faeces of normal consistency. As the cat was still not showing a normal appetite the feeding tube was left in place and enteral nutrition with 200 kcal daily was continued (200 ml Royal Canin Recovery Liquid Dog/Cat per day over five administrations). At home, psyllium husks and lactulose were continued at the same dosage, and the owner was instructed to administer additional nutrition if needed.

Histological evaluation of the rectal mass revealed a submucosal multinodular inflammatory process, which was characterised by anastomosing trabeculae of collagen intermingled with proliferating myofibroblasts, large numbers of eosinophils, macrophages and neutrophils, and fewer lymphocytes, plasma cells and mast cells, surrounding multiple foci of lytic tissue with accumulations of neutrophils, cellular debris and mucus. The process focally reached the mucosa, resulting in focal mucosal ulceration (Figure 5). These findings were consistent with feline gastrointestinal eosinophilic sclerosing fibroplasia (FGESF), and margin evaluation confirmed complete excision

During the first postoperative year regular telephone follow-ups were performed. At all times, the patient had a very good general condition, with normal appetite,

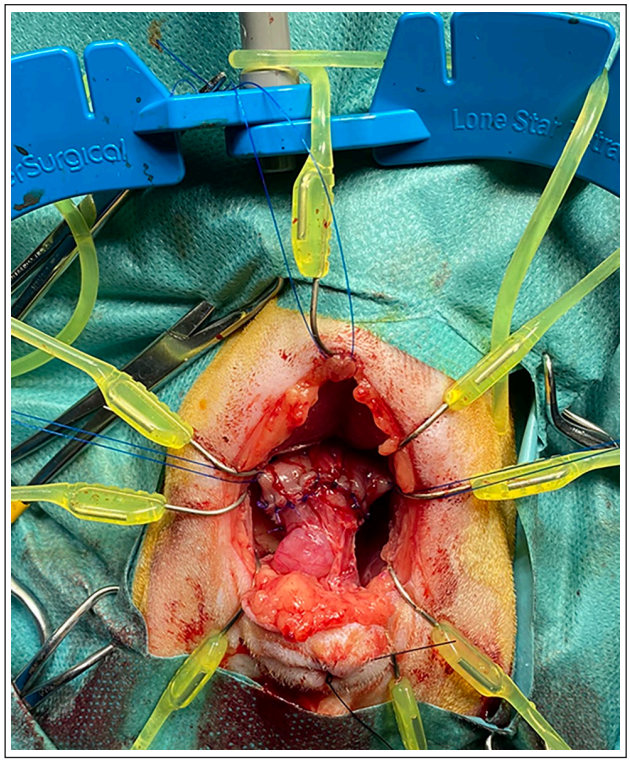

Figure 4 Intraoperative view 3. Close-up of the finished rectal anastomosis

drinking and defecation, and playful behaviour. Fourteen months postoperatively, the cat was presented due to recurrence of dyschezia. Further diagnostic steps, including a recheck CT, revealed the recurrence of a rectal mass. Owing to clinical deterioration and rejection of revision surgery or long-term immunosuppressive treatment, the owner declined further treatment and decided on euthanasia. Further evaluation with cytology, histopathology or necropsy was also declined.

Discussion

FGESF is an inflammatory, non-neoplastic condition with a worldwide distribution of cases in feline patients.[1,2] Previous reports suggested an over-representation in male cats and in Ragdolls.[1] Patient age ranges between 14 weeks and 16 years (median 7–8 years).[2] Described clinical signs are chronic vomiting, diarrhoea or both,[1,3] abdominal pain, weight loss, anorexia and lethargy.[4] The patient presented herein was also a middle-aged male cat with weight loss and anorexia. However, owing to the anatomical location of the mass in this report, the main clinical sign was dyschezia. This clinical sign has not been described previously in the context of FGESF.

In previous reports, FGESF lesions most often occurred at the pyloric sphincter, ileocaecocolic junction or colon.[1,2] Additionally, other locations such as the

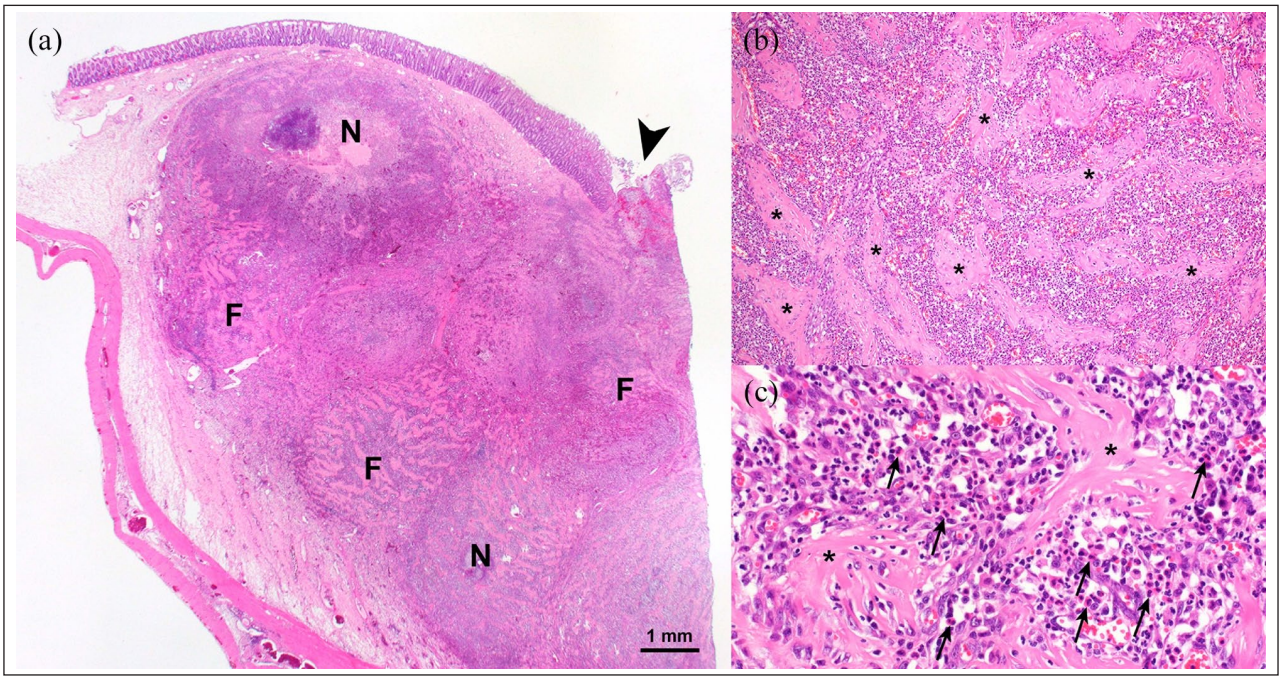

Figure 5 Histology. Photomicrographs of the histological specimen. (a) In the overview, a well-demarcated submucosal mass comprising areas of fibroplasia (F) and necrosis (N) was evident. There was focal ulceration of the overlying mucosa (arrowhead). (b) The fibroplasia was characterised by anastomosing and branching trabeculae of dense collagen (asterisk) surrounded by a highly cellular mixed infiltrate. (c) The infiltrate surrounding the collagen (asterisks) contained numerous eosinophils (arrows), spindle-shaped myofibroblasts and fewer macrophages, neutrophils and lymphocytes

mesentery,[5] retroperitoneum,[6] pancreas[2] and various lymphatic tissues[2,3] have been described. To our knowledge, this case is the first report of rectal FGESF. It is especially important to emphasise that FGESF must be considered as a differential diagnosis when treating cats with a rectal mass. Owners must be informed that this entity may have a more favourable outcome than rectal neoplasia.

The usual pathological appearance is an extensively ulcerated intramural mass potentially associated with enlarged regional lymph nodes.[2] Lesions may be either transmural or affect only the inner layers of the gastrointestinal wall. In our case, this characteristic picture was present mainly in the submucosal layer of the rectum, while there was only minimal mucosal involvement and ulceration, and no involvement of the muscular layer.

Several aetiologies for FGESF have been discussed. It has been hypothesised that cats with a genetic predisposition develop FGESF as a reaction to external pathogens. In particular, the role of bacterial infection is unclear, as bacteria have been found in several cases of FGESF.[1,2,6] However, there have also been cases that lacked bacteria in the lesion.[4,5] Additionally, intralesional fungi have been described in cats,[7] and a similar pathology was found in pumas (*Puma concolor*) associated with intestinal nematodes.[8] In our case, no bacterial or fungal agents were seen with periodic acid–Schiff and Gram stains applied to the histological specimen. One possible explanation for the absence of bacteria could be the antibiotic pretreatment of the patient, although we speculate that the penetration capability of antibiotics into a FGESF lesion may be poor.

Up to 58% of patients with FGESF reported in the current literature showed a systemic eosinophilia, suggesting a potential, unusual presentation of feline eosinophilic granuloma complex.[4,7,9,10] However, our case, and cats reported elsewhere,[1,10] did not show systemic eosinophilia, indicating that a lack of systemic eosinophilia does not rule out FGESF. Furthermore, it is unclear whether the development of systemic eosinophilia is a negative prognostic factor for FGESF. Various studies have reported that eosinophilia resolved under immunosuppressive therapy,[1,5] which may indicate that systemic eosinophilia is a sign of disease progression. Subsequent clinical reports with a higher patient number, standardised work-up and therapy, and long-term follow-up are needed to further evaluate this theory. The patient presented herein also showed a markedly increased creatine kinase (CK). CK is an enzyme expressed by various tissues like the skeletal muscles, the brain and the retina. In the body, it catalyses enzymatic reactions that create adenosine diphosphate/triphosphate, which are important substances in energy balance.[11] Clinically, CK is assayed as a marker of CK-rich tissue damage. The reason for its elevation in our patient is unclear. However, anorectic cats show rapid muscle loss, which can lead to an increase in CK levels. Thus, the anorexia and weight loss in the patient presented here could explain the increased enzyme assay.[11] Interestingly, elevated CK levels were not seen in previously reported cases.[1–13]

Another important aspect of FGESF is the therapeutic approach. Currently, multimodal therapy combining surgical debulking of the mass (biopsy vs complete surgical resection), symptomatic therapy, antibiotic therapy (especially if intralesional bacteria are found) and immunosuppressive therapy using prednisone or ciclosporin A has been described in the literature.[1,5,6] It is suggested that animals treated with immunosuppressive therapy have a longer survival time than those treated without.[1,2] Additionally, a case report presented full remission of a FGESF recurrence in the duodenum using immunosuppressive treatment.[12] In the case presented here, only surgical resection of the mass and symptomatic therapy were used, as the patient showed excellent general health for 14 months and the owner declined long-term medication. However, recurrence leading to euthanasia occurred 14 months after initial surgery. With our experience of the case reported herein, it is of utmost importance to sensitise owners to consider every slight change in the form of faeces or the capacity to defecate as a possible warning signal for recurrent disease. It is unclear whether the use of immunosuppressive therapy would have prevented such recurrence or changed the outcome in our patient. Further studies comparing the two approaches will be needed to further evaluate this aspect.

Importantly, in none of the cases reported in the literature was cytology alone diagnostic for FGESF. Consequently, at the time of decision-making concerning therapy (surgical treatment vs conservative therapy vs euthanasia) the clinician does not usually have an accurate diagnosis. Hence, FGESF should be considered as a differential diagnosis for any mass effect in feline patients, especially in the case of concurrent systemic eosinophilia or multiple eosinophils in cytology.

It may be argued that the caudal mesenteric lymph nodes could have been surgically excised at the time of the rectal mass removal. Although cytology of the lymph node did not confirm a metastatic-like progression of the FGESF in the lymph node, this cannot be excluded with fine-needle aspiration alone. Furthermore, as lymph node involvement is possible in FGESF,[2] a complete resection of the lymph node may have led to further information about the stage of the disease. However, removal of the colonic lymphatic tissue would only have been possible via an abdominal or laparoscopic approach, which would have entailed an additional surgical approach and prolongation of the

surgical time. As at the time of surgery there was no definite diagnosis regarding the lymph node and rectal mass, these supplementary risks determined the decision against this surgical procedure. However, it is important to underline the importance of complete staging in all patients where metastatic disease is possible or assumed.

Retrospectively, it must be mentioned that our patient was not treated with immunosuppressive therapy, although the current literature seems to outline its beneficial effects[13] and even possible remission of FGESF lesions.[12] During the therapeutic course of the case the possibility of immunosuppressive therapy was discussed among the treating veterinary team and with the owner. As the cat initially showed excellent health and as the owner declined long-term medical treatment, where refrained from this therapeutic option.

Conclusions

Our report adds a location and clinical presentation of FGESF to the literature. Additionally, it indicates that patients can have a normal quality of life for several months without the use of long-term immunosuppressive or antibiotic therapy if complete surgical resection of the FGESF is possible. However, immunosuppressive therapy might have prolonged survival or prevented recurrence.

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Ethical approval The work described in this manuscript involved the use of non-experimental (owned or unowned) animals. Established internationally recognised high standards ('best practice') of veterinary clinical care for the individual patient were always followed and/or this work involved the use of cadavers. Ethical approval from a committee was therefore not specifically required for publication in *JFMS Open Reports*. Although not required, where ethical approval was still obtained it is stated in the manuscript.

Informed consent Informed consent (verbal or written) was obtained from the owner or legal custodian of all animal(s) described in this work (experimental or non-experimental animals, including cadavers) for all procedure(s) undertaken (prospective or retrospective studies). No animals or people are identifiable within this publication, and therefore additional informed consent for publication was not required.

ORCID iD Laura M Goffart https://orcid.org/0000-0002-9135-356X
Simona Vincenti https://orcid.org/0000-0003-3583-215X

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