LETTERS TO THE EDITOR
EDİTÖRE MEKTUPLAR
A case of polyarteritis nodosa diagnosed after recurrent, multiple intestinal perforations

Tekrarlayan çoğul intestinal perforasyon sonrası tanı konan poliarteritis nodoza olgusu

To the Editor

A 44-year-old male was admitted due to recurrent fever, abdominal pain, chronic diarrhea, and weight loss lasting for six months. On physical examination, he was subfebrile with normal vital signs but no arthritis, arthralgias, skin rash, or oral aphthae, and the remainder of the examination was normal.

His complete blood count was hematocrit 35%, leukocytes 16000/mm^3, and platelets 526000/mm^3, with marked eosinophilia (25%). The erythrocyte sedimentation rate (ESR) was 105 mm/h and C-reactive protein (CRP) level was 156.9 mg/L. His complete blood chemistry was normal but revealed a polyclonal gammopathy, and the immunoglobulin E level was 375 IU/ml (< 80). The urinalysis was normal and a chest X-ray and plain abdominal radiograph revealed no pathology. Repeated Ziehl-Neelsen stains, polymerase chain reaction assays, and cultures of sputum and feces remained negative for tuberculosis, and microscopic examination of the feces revealed no ova or parasites. Computerized tomographies of the thorax and abdomen also revealed no pathology.

A colonoscopy was performed, and patchy mucosal edema and hyperemia in the presence of a few scattered erosions were noted; histopathology disclosed severely active eosinophilic infiltrations. The patient was thought to have eosinophilic colitis and placed on steroid treatment (methylprednisolone 80 mg/d, i.v.) with a dramatic clinical and biochemical response. However, on the 8th day of treatment, the patient developed ileal perforation and underwent a laparotomy with closure of the perforation, but he experienced recurrent perforations requiring reoperation two more times in three days, ending with a segmental ileal resection, anastomosis, and loop ileostomy.

In search of recurrent perforations, ophthalmoscopy and skin pathergy test were normal and blood, stool, and urine cultures remained negative for typhoid fever. All three resection materials were reviewed together and the patient was diagnosed to have disseminated necrotizing transmural arteritis complicated by secondary intestinal ischemia and perforation (Figure 1).

HBs Ag, anti-HCV, VDRL, cryoglobulins, perinuclear, cytoplasmic, and MPO-ANCA were found within normal limits. Rheumatoid factor level was 386 IU/ml (<15), complement levels were low (0.53

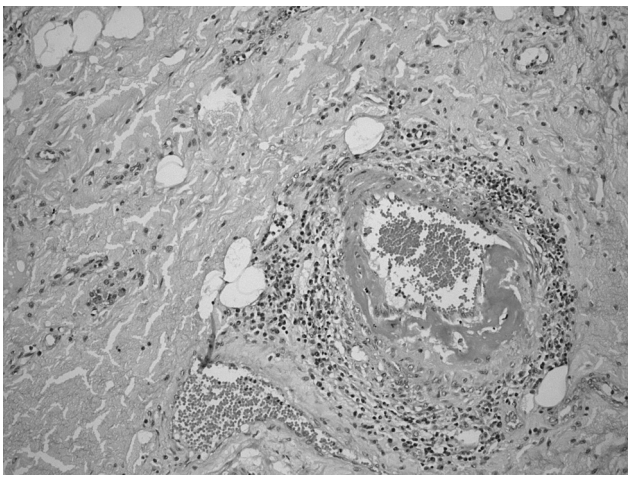

Figure 1. Under high power, medium-sized arteries located in submucosa and subserosa showed abundant fibrinoid necrosis, luminal narrowing and lymphocytic inflammation (hematoxylin and eosin, x200).

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and 0.05 IU/ml for C_3 and C_4, respectively), and ANA was slightly positive. Electromyography of the extremities revealed asymmetric, mild sensorial polyneuropathy, but a transthoracal echocardiography and mesenteric angiography were normal.

The patient was diagnosed to have polyarteritis nodosa (PAN), and cyclophosphamide 1 g/q 4 weeks i.v. was started in addition to his steroid treatment. The steroid dosage was then gradually tapered and cyclophosphamide perfusions were stopped after nine months. Currently, the patient is receiving only methylprednisolone 5 mg/d and has been under our close follow-up without any further problems for more than two years.

In PAN, the gastrointestinal tract is involved in approximately half of the patients (1-2), significantly lowering the survival rates particularly in those with perforations (2-4). However, the demonstration of vasculitis in endoscopic biopsies of the gastrointestinal tract is difficult as they are mainly composed of mucosa and the blood vessels in the mucosa are mainly capillaries (5, 6). Thus, in our case, PAN was diagnosed after histopathological examination of surgical specimens showing transmural necrotizing vasculitis. In the differential diagnosis, Churg-Strauss syndrome was eliminated due to the absence of allergic rhinitis, asthma, granulomatous vasculitis, and hypereosinophilic syndrome via typical histopathological findings of PAN.

Challenging symptoms like recurrent abdominal pain, diarrhea, fever, weight loss and recurrent, multiple intestinal perforations should lead physicians to suspect vasculitis since early deaths are reported as a result of insufficient or inappropriate therapy.

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Gastric outlet syndrome caused by persimmon (Diospyros kaki) bezoars

Trabzon hurmasına (Diospyros kaki) bağlı gelişen bezoarların neden olduğu mide çıkış sendromu

To the Editor

Bezoars are collections of indigestible foreign material that accumulate and coalesce in the gastrointestinal tract, usually the stomach. Phytobezoars are the most common type of bezoars, compo-

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sed of vegetable matters such as celery, pumpkin, grape skins, prunes, raisins, and most notably, persimmon (1). Persimmon (Diospyros kaki) is a fleshy fibrous tropical fruit produced and consumed in various countries, including Turkey (2, 3). We describe herein two cases of phytobezoars caused by persimmon consumption who presented with gastric outlet syndrome.

A 50-year-old male patient presented with vomiting after meals. He also had abdominal pain and nausea for three weeks. On physical examination, epigastric tenderness and clapotage were determined. He had a history of type 2 diabetes mellitus for 10 years and of eating unpeeled persimmons with seeds. Endoscopic examination showed bezoar in the antrum passing through the pylorus to the duodenum. The bezoar was hard and composed mostly persimmon seeds.

A 45-year-old female patient admitted to the hospital with vomiting for two weeks. She had abdominal pain, early satiety and weight loss. On physical examination, there was only abdominal tenderness. She had a history of eating unpeeled persimmons with seeds and of multinodular thyroid surgery, but she was euthyroid on admission. Endoscopic examination revealed the bezoar in the antrum closing the pylorus (Figure 1).

The main risk factors for bezoars are abnormal mastication, vegetarian diet, gastric operation, diabetic gastroparesis, hypothyroidism, and ingestion of foods like persimmons. Many patients with bezoars have symptoms such as feelings of epigastric distress, nausea, vomiting, anorexia, early satiety, and weight loss, like in our cases. In the male patient's history, consumption of unpeeled persimmons with seeds and type 2 diabetes mellitus were the etiologic factors. He had been eating a plateful of unpeeled persimmons with seeds every night after dinner for six weeks. In the female patient's history, there was no evidence of risk factors but she had symptoms of early satiety and abdominal discomfort for a long time, as in patients with dysmotility-like dyspepsia. She had been eating a plateful of unpeeled persimmons with seeds daily for two months before admission to our hospital.

In our cases, phytobezoars did not respond to endoscopic therapy. Surgical removal of bezoars was performed. Foods like persimmon contain large amounts of nondigestible dietary fiber such as cellulose, hemicelluose, lignin and tannin. The persimmon is characterized by its high level of tannic acid (tannins) (4). Tannins in high concentrations form a coagulum when exposed to gastric acid. When sufficient quantity of persimmon fruit accumulates, phytobezoars may develop (5). Benharroch et al. (6) reported that while ingestion of persimmon carried a 9.8-fold risk of bezoar development, ingestion of the unpeeled fruit increased the risk of this complication 56 times over that of age- and sex-matched controls. After diagnosis and localization of the bezoar, treatment involves removal and prevention of recurrence.

In conclusion, we recommend that patients with bezoar history, prior gastric surgery, and comorbid illnesses such as diabetes or end-stage renal diseases on dialysis should be warned particularly against eating foods like persimmons with seeds, especially in their unpeeled forms.

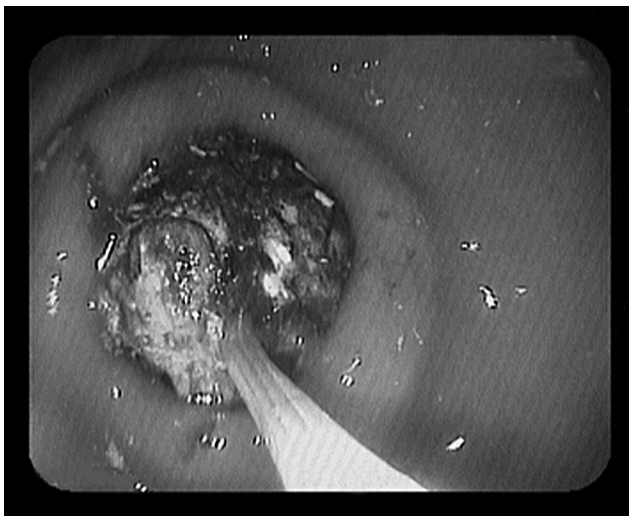

Figure 1. Phytobezoar in the antrum obstructing the pyloric channel.

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Jejunogastric intussusception: a rare cause of hematemesis

Jejunogastrik intusussepsiyon: Hematemezin nadir bir nedeni

To the Editor

Intestinal intussusception, although common in the pediatric population, is rare in adults. Adult intussusception is associated with a definable pathologic lesion or lead point in 70%-90% of cases, and about 40% of them are caused by a primary or secondary malignant neoplasm (1-3). Jejunogastric intussusception (JGI) is a rare complication of gastrectomy and gastrojejunostomy. In acute JGI, the clinical findings are severe and the prognosis is poor. The mortality rates rise from 10% to 50% if the delay in diagnosis is more than 48 hours after the onset of symptoms (4, 5). Therefore, early diagnosis of JGI is important.

A 31-year-old man was admitted to the emergency department for severe colicky pain that began in the epigastrium 24 hours prior to admission and gradually extended to the abdomen, followed by bilious vomiting and subsequent hematemesis. He had history of vagotomy and gastrojejunostomy carried out 16 years before because of a bleeding duodenal ulcer. On admission, laboratory studies showed a white blood cell count (WBC) of 19,600/µL, and a serum hemoglobin (Hb) level of 16.4 g/dl. Upper endoscopy was performed. There was no optimal view because of large blood clots. Hematinized blood continued to drain from the nasogastric tube at follow-up, and blood counts showed WBC of 21,500/µL and Hb of 10.1 g/dl. Follow-up endoscopy revealed a lobulated, bluish-brown, congestive mass with petechial hemorrhage that occupied the entire body of the stomach (Figure 1). Abdominal CT demonstrated intestinal loops ex-

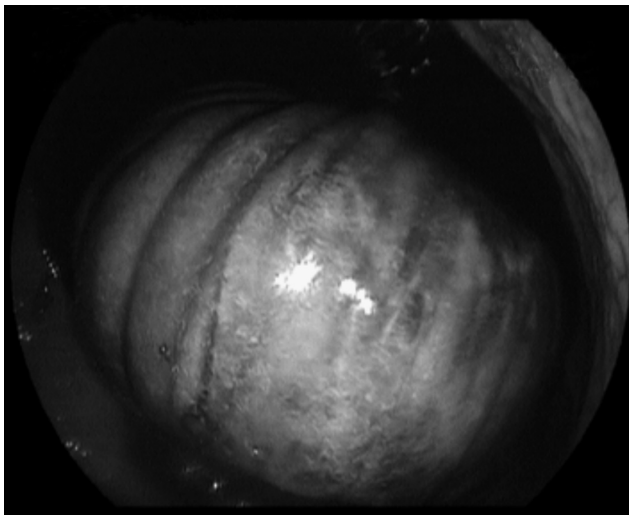

Figure 1. Endoscopic appearance of the intussuscepted jejunal segment in the body of the stomach.

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tending into the gastric lumen through the posterior wall of the stomach, with thickened valvulae conniventes, and presence of minimal contrast material and air at the intestinal loops (Figure 2). Because these findings suggested necrosis of the bowel wall, the patient was operated and a loop appeared congested and ischemic when disclosed. The ischemic segment was resected. The clinical picture of the patient improved during the follow-up.

Intussusception most commonly occurs in childhood and is rare in adults, accounting for only 5% of all cases of intussusception (6). JGI is an uncommon complication of gastrointestinal surgery. Only 16 cases of JGI were observed during 1907–1980 according to results from the Mayo Clinic (4, 7). On upper endoscopy, it is important to visualize the anastomosis and the intestinal loops intussuscepted through it to the stomach. However, it may not be easy to distinguish the congestive, bluish-brown mass in a stomach filled with blood and clots. Computerized tomography (CT) is quite valuable in imaging intra-abdominal pathologies, and may reveal the intestinal loop intussuscepted to the stomach in detail. Definitive diagnosis was established on abdominal CT performed in order to clarify the acute abdomen, and emergency surgery was carried out.

In conclusion, JGI should be considered in all patients with a history of gastrectomy who present with colicky abdominal pain, vomiting and hematemesis. Abdominal CT is quite valuable in the diagnosis.

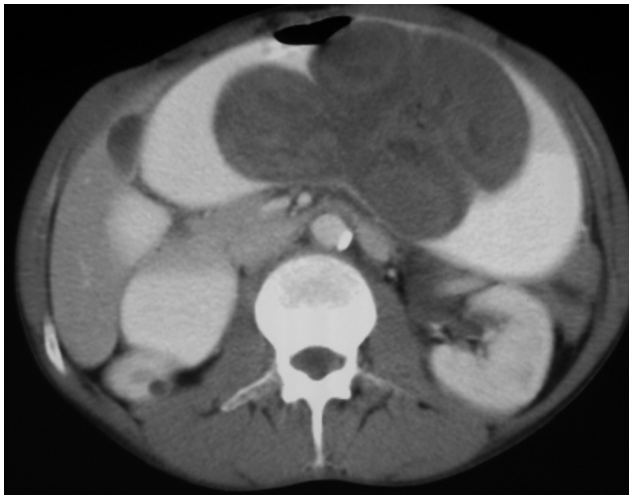

Figure 2. Abdominal CT showing jejunogastric intussusception.

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A case report of intestinal obstruction due to phytobezoar within Meckel's diverticulum

Meckel divertikülü içindeki fitobezoara bağlı oluşmuş bir intestinal obstrüksiyon olgusu

To the Editor

Meckel's diverticulum is the most common congenital anomaly of the gastrointestinal tract and may present with obstruction due to intussusception, volvulus around an associated fibrous or omphalomesenteric band, adhesion from an inflammatory process, or incarceration within hernia sac (1). In this report, we present a case of obstruction of the small intestine due to phytobezoar formation in Meckel's diverticulum, which is an exceedingly rare condition. To the best of our knowledge, there are only eight cases in the English literature (2, 3).

A 56-year-old man admitted to the emergency service with a two-day history of nausea, vomiting, abdominal pain and distension. He had been evaluated several times in outpatient clinics for intermittent episodes of abdominal pain and nausea for the last two years but there had been no specific pathological finding. He had no abdominal operation history and his upper gastrointestinal tract endoscopy and colonoscopy performed two months before were found to be normal. His physical examination revealed a distended abdomen with no peritoneal irritation signs like direct/indirect tenderness. Routine laboratory blood tests were normal. An abdominopelvic contrast-enhanced computed tomography (CT) was carried out and revealed non-specific multiple dilated low attenuation fluid-filled loops of the small bowel. A nasogastric suction and intravenous fluid therapy were commenced, but his condition deteriorated slightly through acute abdomen that necessitated diagnostic laparotomy. A Meckel's diverticulum that was obstructed by a bezoar approximately 100 cm proximal to the ileocecal valve was observed intraoperatively and resected without any complications (Figure 1). He was discharged from the hospital after three days with no complications.

The main risk factors for phytobezoar formation are abnormal mastication, vegetarian diet, ingestion of persimmons, gastric operation, diabetes, gastroparesis, and hypothyroidism (4). Overall incidence of bezoar-induced small bowel obstruction is relatively rare (4.8%), but has life-threatening complications like ulceration, perforation, intussusception and obstruction, with a mortality rate of 30% of all cases (5, 6). Preoperative diagnosis is difficult; inconsistent with our case, in which CT revealed no specific signs of bezoar formation, CT scan findings were previously reported to be helpful (7). Surgical management entails milking the offending object into the cecum or performing enterotomy for retrieval in difficult cases. Obstructive Meckel's diverticulum with phytobezoar formation is uncommon but should always be kept in mind in the differential diagnosis of an acute abdomen.

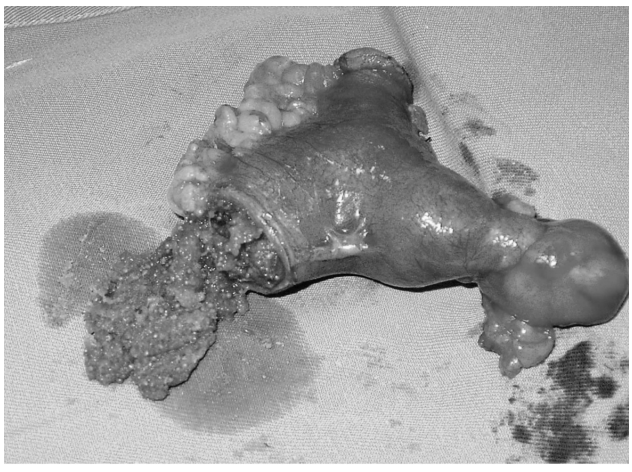

Figure 1. Phytobezoar in Meckel's diverticulum.

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Simultaneous repair of bilateral recurrent femoral hernias complicated with rectus sheath hematoma

Aynı seansta onarılan bilateral nüks femoral herni olgusunda gelişen rektus kılıfı hematomu

To the Editor

Femoral hernias are rare and constitute only 1% of all groin hernias in men. Recurrence may be an important problem after femoral hernia repairs. Recurrence can appear either following inguinal hernia repairs as a missed femoral hernia or as a true recurrence after a previous femoral repair (1-3).

Bleeding complications after groin hernia repairs are not common; most result in wound hematoma/seroma, at a rate of 2-16.4% (4, 5).

Herein, we report a 70-year-old male patient with a painful irreducible lump on the right groin who was admitted to our surgery department. The patient had had two separate inguinal hernia repairs 24 and 14 years ago at different hospitals.

During the operation under general anesthesia, the right side was explored first and a femoral hernia was found without any other type of groin hernia.

The same finding was observed on the left side and the hernia was similarly repaired. The patient was given thromboembolism prophylaxis after the operation with two consecutive doses of 0.2 ml enoxaparin. He was discharged without a wound hematoma on day 2.

The patient returned to the hospital on day 7 with a complaint of painful mass on the lower abdomen. Ultrasound revealed a cystic mass 72x53 mm in diameter with internal septates. The lesion was located between the bladder and the rectum preperitoneally. Computed tomography confirmed this finding and showed a posterior extension to the left side (Figure 1).

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A diagnostic puncture was decided. After aspirating 300 ml hemorrhagic fluid percutaneously, the final diagnosis of rectus sheath hematoma was reached. The hemoglobin level did not show a marked decrease. No recurrence was observed as of the three-month follow-up.

In the presented case, the recurrence on the right side seems to be an overlooked femoral hernia, since it appeared only three days after the first surgery. However, it would be difficult to draw the same conclusion for the left side because of the 14-year interval between the previous repair and the realization date of the recurrence. Furthermore, the operation report from the first repair could not be located, so the possibility of a true femoral recurrence after a previous femoral hernia repair can not be ruled out.

In this particular case, a large rectus sheath hematoma was diagnosed on postoperative day 7. It was probably secondary to the difficult dissection and an unrecognized bleeding from epigastric vessels or within the scar tissue at the vicinity of the rectus sheath. Oral anticoagulant usage and thromboembolism prophylaxis with heparin are also well-documented causes of rectus sheath hematoma. Whatever the etiology, the underlying cause is a vascular injury, which is frequently to the epigastric vessels, or bleeding from a direct tear of the muscle.

The treatment of rectus sheath hematoma is conservative in many cases (6, 7); however, it can require surgical intervention or endovascular embolization in patients who develop clinical deterioration (6). In the present case, an almost complete resolution could be achieved with a conservative approach. Computed tomography after three months displayed a small remainder on the left anterolateral edge of the rectus muscle, but the patient had no complaint.

In conclusion, femoral hernia can be easily missed during groin hernia surgery and in secondary cases may be associated with morbidity.

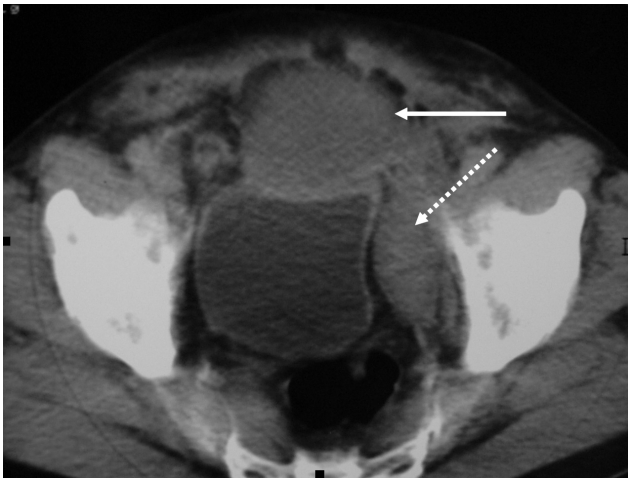

Figure 1. The tomographic features of the case on the day-7. Large rectus sheath hematoma is seen [solid arrow] with its posterior extension to the left side [dotted arrow].

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Pseudomyxoma retroperitonei presenting with a skin fistula

Cilt fistülü ile tanı konulan psödomiksoma retroperitonei

To the Editor

A 51–year-old lady was admitted with a six-month history of purulent and mucinous discharge from the right flank. Over the last six months, she was followed as having a soft tissue infection and given different antibiotic regimens but complete remission could not be achieved. Physical examination revealed a mass on the right flank. An ulcer surrounded by an edematous and erythematous skin was identified on the top of the mass. The serum CA19-9 and CEA concentrations were 47.05 (0-39 U/ml) and 12.2 (0-5) ng/ml, respectively. Abdominopelvic computed tomography scanning demonstrated a well-defined mass (15x11x7 cm) in the right retroperitoneal space, infiltrating the lateral abdominal wall muscles and iliac bone, and extending to the right flank region (Figure 1). Colonoscopic examination was normal.

The patient underwent a laparotomy, which revealed a retrocecal mass infiltrating the lateral abdominal wall muscles and iliac bone. The mass did not involve the colon or other intraperitoneal organs. The retrocecal appendix buried in this mass was determined as the origin neoplasia. An en bloc resection of the mass with appendectomy including a portion of the iliac bone was performed.

The pathological diagnosis was mucinous adenocarcinoma of the appendix. Chemotherapy and radiotherapy were administered after surgery.

Primary adenocarcinoma of the appendix is an extremely rare tumor, and is found in only 0.01-0.2% of all appendectomy specimens. Mucinous cystadenocarcinoma of the appendix is rarer and constitutes 6% of all appendiceal tumors (1).

Progression of these tumors is usually transperitoneal, resulting in pseudomyxoma peritonei. In very rare instances, rupture of the mucocele of the retrocecal appendix to the retroperitoneal space may occur. This extremely rare condition was first described and termed as “pseudomyxoma extraperitonei” by Moran and Morgan in 1988, and later as “pseudomyxoma retroperitonei” by Shelton in 1994. The nonspecific symptoms may lead to a delay in diagnosis. The most common symptoms are sensation of fullness on the right flank, slowly progressing pain, loss of appetite, and fatigue (2).

Our patient presented with a skin fistula, and to our knowledge, there are only two other reports in the medical literature defining the potential of pseudomyxoma retroperitonei to cause fistulae (2, 3).

This large tumor was treated with wide resection of the tumor and appendectomy, and additional adjuvant chemoradiotherapy was administered. Resection of the mass and the appendix may be an alternative to right hemicolectomy when the colon

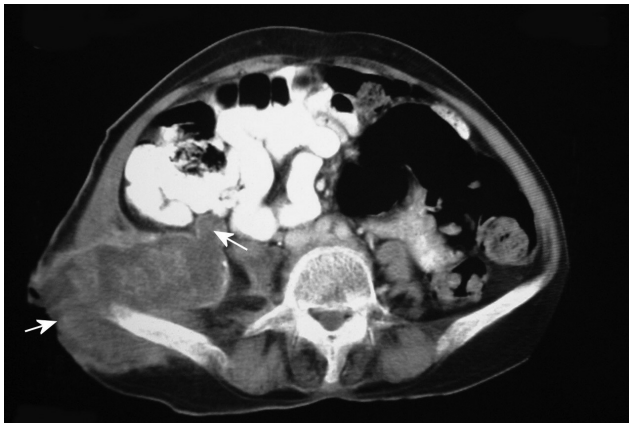

Figure 1. CT demonstrated a well-defined mass in the right retroperitoneal space, infiltrating the lateral abdominal wall muscles and iliac bone and extending to the right flank region with a skin fistula.

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is not infiltrated, but routine chemotherapy and radiotherapy must be the part of the treatment. Our patient has been alive for three years following surgery without any sign of recurrence.

Prognosis of the pseudomyxoma retroperitonei may be better than with pseudomyxoma peritonei as a result of isolation of the tumor in the retroperitoneal region. The skin fistula may have helped to establish a tract that inhibited the growth of the tumor to other intraabdominal sites (2).

In summary, physicians must be aware that an ulcerated skin fistula on the right flank may be the only manifestation of a retroperitoneal pseudomyxoma of the appendix.

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Imaging studies detecting retrorectal tumors

Rektorektal tümörlerin belirlenmesinde görüntüleme yöntemleri

To the Editor,

I praise your journal and the authors Menteş et al. (1) on the excellent publication titled, “Retrorectal tumors: a case series.” The authors note that rectal pain and perirectal mass sensation were the patients’ main symptoms and they accurately note that, "suspicion is the main factor in the diagnoses of these tumors."

The authors correctly declare that the tumors can be detected using computed tomography (CT) and magnetic resonance imaging (MRI). The data from Menteş et al. show that in 6 of 8 cases (75%), the retrorectal tumor was below the level of the fourth sacral vertebrae. Thus, I would humbly add that the proper level being imaged by CT or MRI would be crucial. Specifically, a typical "lumbar” MRI or even a "lumbosacral” MRI would often fail to include images that are low enough (inferior enough) to show a tumor that is anterior to the coccyx or lower sacrum. Imaging the appropriate level may seem obvious. However, I have seen many patients with perirectal pain and/or coccyx pain (coccydynia, coccygodynia, tailbone pain) where the treating physician had ordered a lumbar or lumbosacral MRI, thus failing to include the symptomatic region of the coccyx and lower rectum. The wonderful article by Menteş et al. confirms what I have noted only anecdotally in my own clinical practice, i.e., that such lumbar or lumbosacral imaging levels would often miss these retrorectal tumors.

I thank the authors and this journal for this very educational article. I hope that my comments will further highlight the importance of the authors’ data, which can guide physicians to order the most appropriate imaging studies.

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Reply of the Authors

Dear Editor,

We are most thankful to the appreciation and contributions of Dr. Foye. It is perfectly true that the proper level being imaged by CT or MRI would be crucial and that a typical "lumbar” MRI or even a “lumbosacral” MRI would often fail to include images that are low enough to show a tumor that is anterior to the coccyx or lower sacrum. That’s a point we did not emphasize in our manuscript.

The important point stressed by Dr. Foye, as well as the common mistakes in surgical treatment of retrorectal tumors, stem from the well-known clinical fact that the more unusual and rare a lesion is in clinical practice, the more mistakes are realized in its diagnosis and treatment. That’s why we tried to show in detail the unusual surgical technique of posterior parasagittal approach. Although the posterior approach is very efficient in reaching and treating a low retrorectal tumor, a classical and well-known transabdominal approach will complicate good exposure.

We hope that clinicians will benefit from the details involved in our case series, as well as the recommendations of Dr. Foye, if any one of them is unlucky to meet one of these rare tumors.

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