INTRODUCTION

Crohn’s disease is a chronic, relapsing inflammatory disorder characterized by transmural mucosal inflammation of any segment of the gastrointestinal tract that may be complicated by fibrosis, obstruction, and sinus tracts that can penetrate the serosa and cause microperforations and fistulæ. It affects up to 480,000 people in the United States. Given its usual presentation in young adulthood, most patients suffer from disease complications throughout the majority of their working lives, decreasing both productivity and quality of life. Estimated healthcare costs per year exceed $12,000 in medical services and drug treatments, with the majority of costs arising from inpatient hospitalizations (1).

We report a case of a young woman who was mistakenly diagnosed with Crohn’s disease based on her presentation of a persistent rectovaginal fistula. With continued and failed medical and surgical therapies, amounting to total healthcare costs exceeding $135,000, the patient was ultimately discovered to have factitious disorder, manifested by self-inflicted wounds. Our case illustrates the importance of early diagnosis of this frequently missed disorder in an effort to avoid unnecessary and potentially harmful procedures and treatments for other mistakenly diagnosed disease entities and to initiate early and essential treatment for psychiatric illness.

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Fistulizing Crohn’s Disease

A CASE TO REMEMBER

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CASE PRESENTATION

A 23-year-old female who had recently undergone appendectomy and partial colectomy for appendicitis presented two months postoperatively with three weeks of rectal bleeding and right-sided abdominal pain. Her past medical history included an uncomplicated vaginal delivery eight years previously, menorrhagia, gastroesophageal reflux disease, cholecystectomy, and recent appendectomy. She did not take medications and had no allergies. As a single mother of an eight-year-old daughter, she was living at home with her parents. She had a 10-pack-year tobacco history and denied alcohol consumption. Her family history was noncontributory.

Computed tomography (CT) scan of the abdomen and pelvis was normal, except for postoperative changes in the right lower quadrant. Pelvic exam revealed a large fistula communicating with the rectum. Subsequent colonoscopy confirmed a large rectovaginal fistula with significant inflammation in the distal rectum and normal colonic mucosa proximally (Figure 1). Proctosigmoid and vaginal biopsies revealed marked inflammation and ulceration, but without the presence of granulomas or malignant changes. She was then given a presumed diagnosis of Crohn’s disease and underwent an uncomplicated laparoscopic diverting sigmoid loop colostomy with Hartmann’s pouch to promote fistula healing.

Over the next three months, the patient had multiple episodes of recurrent rectal bleeding. Because of the non-healing rectovaginal fistula and aggressive medical and surgical therapy, she subsequently underwent rectovaginal fistula closure with muscle flap. Postoperatively, the patient did well for two months until she developed recurrent rectal bleeding. A gastrograffin enema revealed the recurrence of a rectovaginal fistula, which was then treated with a prolonged course of antibiotics.

Three months later, she was rehospitalized with stomal bleeding. Endoscopic evaluation revealed a localized area 2 cm from the stoma with deep serpiginous ulcers and blood clots. Biopsies throughout the colon and terminal ileum were otherwise normal. Stomal bleeding continued, in spite of medical and endoscopic therapies, requiring multiple blood transfusions. A second endoscopic evaluation via the stoma revealed worsening ulceration, which culminated in an explorative laparotomy with partial sigmoid colectomy and end-colostomy reconstruction. The pathology result of the resected sigmoid colon revealed mucosal ulceration, fissuring, and transmural inflammation without the presence of granulomas or evidence of crypt destruction (Figures 2A–2C). These nonspecific findings raised the possibility of alternative diagnoses.

Four weeks later she was readmitted with abdominal pain, without laboratory or radiographic abnormalities. On day two of her hospitalization, the patient became increasingly hostile and noncompliant, refusing further blood draws and diet advancement. She was insistent on having a total colectomy. The following day, she developed severe stomal bleeding, requiring four units of blood and transfer to the intensive care unit with the possibility of surgical intervention. Repeat endoscopy revealed a large ulcer with a bleeding visible vessel just proximal to the stoma. Hemostasis was successfully achieved with endoscopic clipping. With control of bleeding precluding the need for surgery, the patient became immediately agitated and angry, attempting to pull her IV lines and ostomy bag and threatening to leave the hospital against medical advice.

Psychiatry was consulted the next day. She admitted to the ongoing self-infliction of her wounds with a needle through the vagina, rectum, and stoma, since her initial presentation. She was diagnosed with factitious disorder and ultimately transferred to the inpatient psychiatric unit for supportive psychotherapy.
DISCUSSION

Diagnosing factitious disorder (FD) by the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM IV) criteria requires two clinical features: deception or the intentional production or feigning of symptoms, and a psychological motivation to assume the role of the sick patient without any evidence of external incentives. There are subsets of the disorder—those with predominantly psychological signs and symptoms, and those with predominantly physical signs and symptoms. A third subset includes both psychological and physical signs and symptoms. Another classification system, the International Classification of Diseases, Tenth Revision (ICD-10), defines factitious disorder as the intentional production or feigning of symptoms or disabilities, either physical or psychological (2). Munchausen’s syndrome is a form of FD reserved for the most severe and extreme cases, and accounts for only 10% to 20% of patients with factitious disorders (2). It is named for Baron von Munchausen, a German cavalry captain, who was famous for his unbelievable adventure stories (3).

The exact incidence of FD is unknown, and the bulk of published data on this disorder are anecdotal case reports. In 1,538 patients on a neurology ward with feigning neurological symptoms, Bauer and Boeg-ner diagnosed 0.3% with factitious disorder in one year. Sutherland and Rodin evaluated 1,288 patients referred for psychiatric consultation, and labeled 0.8% with factitious disorder (2). While the prevalence of factitious

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disorder is difficult to ascertain, it is believed that this illness is still vastly underreported (3).

There are some common features found in patients diagnosed with FD. The typical patient is a young woman who is involved with health care. Working in the medical field provides these patients with the knowledge of how disease may be produced artificially and allows access to equipment, such as chemicals and syringes that can be used to produce symptoms or create a medical condition (3). One study from 1983 performed over 10 years, found that in 41 patients with factitious disorder, 95% were female, the average age was 33, and 68% held jobs in the health care field. In another study by Carney and Brown, out of 43 patients, 76% were female, the mean age was 34-years-old, and 50% were involved in the health care field (4). The more common presenting complaints of factitious illness are listed in Table 1 (5). A patient with a true underlying medical condition could also develop FD, as they gain more medical knowledge about their disease process, and use it to feign a medical condition (6). Within the patient’s history, there may be elicitable evidence of emotional neglect or emotional, physical, or sexual abuse (3). Some speculate that feigning a medical condition may represent an opportunity to escape from recent stressful events in the patient’s life (3,4). The detection of FD is challenging and often hampered by the natural tendency of health care providers to believe that patients’ symptoms are real. There are clinical clues, however, that may raise suspicion for a factitious illness, as listed in Table 2 (5).

The financial repercussions of this illness cannot be overstated. Medical resources are expended at an alarming cost with frequent hospitalizations, and invasive procedures (7). In a case report from 1985, one woman incurred $46,000 dollars in medical bills in one year, and $500,000 dollars in her lifetime, by repeatedly presenting with factitious illnesses (8). Part of the issue with the cost of this illness is the ease with which these patients can receive medical care. They often present to multiple hospitals, give skewed and incomplete medical histories, and limit access to family members who could offer more history. Even if a psychiatric diagnosis is given to a patient, physicians are still likely to explore causes of their medical complaint due to concerns for real concomitant medical disease and fear of being charged with malpractice (8).

A patient with factitious disorder is not only difficult to diagnose, but even more difficult to treat. A majority of patients with factitious disorder refuse treatment (9). When this illness was first discovered, there were early discussions on limiting these patients’ access to medical care, in the form of “Black Lists” to identify these patients (3). Within the literature, there is controversy regarding approach to treatment, specifically whether or not confrontation with the patient about their self-induced complaints is beneficial. In a study by Krahm, et al, 93 individuals with factitious dis-

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**Table 1**

Common Presentations of Factitious Disorder in Patients

- Hypoglycemia (Insulin induced)
- Fever (Self Induced)
- Dyspnea
- Dramatic Bleeding and Hematologic Disorders
- Dermatitis, Recurrent Skin Infections (Self inflicted)
- Neurologic Disorders (Seizures)
- Flank Pain and Hematuria
- Cardiac Symptoms (Chest pain)
- Gastrointestinal Manifestations (Abdominal pain, Diarrhea, Vomiting, Gastrointestinal Hemorrhage)

**Table 2**

Clinical Clues That Raise Suspicion for the Diagnosis of Factitious Disorder

- Dramatic or atypical presentation
- Elusive medical history with vague and inconsistent details
- Willingness and eagerness to undergo physical evaluation, treatment, and procedures, with a calm acceptance of the associated risks and discomfort
- Extensive medical history which includes multiple hospitalizations
- A familiarity with medical terms and procedures and knowledge of textbook definitions of illness
findings, endoscopic disease, histopathology, and laboratory and radiographic abnormalities. Relying on only a portion of these parameters may hastily lead to an erroneous diagnosis of inflammatory bowel disease. This case also reminds us of the importance of comprehensive history-taking, to identify factors that may raise suspicion for feigned illness.

Literature on factitious disorder is clearly lacking, with a majority of available data coming from case reports. Given litigious concerns, physicians are less likely to consider factitious disorder, until all other medical diagnoses are explored and clearly ruled out. This climate enables these individuals who feign medical conditions to continue their course, undergoing unnecessary and potentially harmful tests and treatments for self-inflicted medical disease, instead of getting essential treatment for their psychiatric illness. Our case illustrates a very real and underreported condition and serves to highlight the importance of early diagnosis of factitious disorder.

CONCLUSION

The tremendous impact of factitious disorder on the patient, family, and health care system is unquestionable. Our patient accumulated medical expenses upwards of $135,553 over the course of 18 months, amounting to eight discrete hospitalizations.

In retrospect, a review of our patient’s pathology slides from early biopsies of the peri-fistula area revealed a greater involvement of inflammation in the serosal layer of the rectum, with less tissue damage mucosally, suggesting extrinsic rectal trauma through the vagina (Figure 2c). Unfortunately, given the transmural extent of inflammation, a mistaken diagnosis of Crohn’s disease was given, resulting in three unnecessary surgeries. It was only during her final hospitalization, when the patient displayed controlling, hostile, and disruptive behavior and her condition took a fluctuating clinical course, including the rapid and dramatic development of new complications at a time when she was otherwise stable, that we entertained the possibility she was feigning her illness. Crohn’s disease is a diagnosis based on a combination of physical

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References