CASE PRESENTATION

An 16 year-old male patient presented with an eight-month history of chronic constipation, and hematochezia. There was no history of abdominal pain, tenesmus, decreased appetite, weight loss or diarrhea. Physical examination and routine laboratory studies were non-contributory. Colonoscopy revealed a polypoid ulcerated mass 5 cm from the anal verge (Figure 1). Histology is as shown (Figure 2).

Most likely etiology of the rectal bleeding in this patient is:

1. Rectal carcinoma
2. Rectal polyp
3. Solitary rectal ulcer syndrome (SRUS)?

The patient was diagnosed with SRUS and treated conservatively with a high fiber diet and laxatives. Clinical course was complicated by persistent symptoms and rectal prolapse that required surgical excision and a rectopexy.

Solitary rectal ulcer syndrome is a chronic benign inflammatory disorder that predominantly affects young adult females.1 Pediatric literature is limited to case reports and studies with varying treatment protocols and outcomes. The term SRUS can be misleading as ulcers occur only in one fourth of the adults with the remainder having non-ulcer lesions. These lesions may be multiple and circumferential with a polypoid, plaque-like or ulcerated appearance on endoscopy.1, 2 The

Figure 1.

Figure 2.
polypoid variant of SRUS is very rare and may mimic inflammatory bowel disease, polyps, or carcinoma of the rectum. Hence misdiagnosis or delay in diagnosis is common. Clinical presentation may include constipation, rectal bleeding, straining with defecation, incomplete evacuation of feces, self-digitation and rectal prolapse. The etiology remains obscure and may be secondary to excessive straining efforts that force the anterior rectal mucosa into the anal canal causing strangulation, edema, and ulceration. Histology is diagnostic with fibromuscular obliteration of the lamina propria and disorientation of muscle fibers.

Initial management is conservative and focused on treating the underlying constipation and behavioral measures to avoid straining. Behavioral modification remains challenging in pediatric practice. Sulfasalazine, steroid and sucralfate enemas have been used with initial improvement of symptoms. However recurrence is common. Argon plasma coagulation has been shown to promote ulcer healing and control bleeding in adults. Surgery is indicated in patient’s refractory to medical therapy and includes rectopexy, local excision of the ulcer and rarely colostomy.

Though there are increasing number of cases in the pediatric literature, misdiagnosis and a delay in diagnosis remains common. We emphasize the importance of maintaining a high degree of suspicion in children who present with rectal bleeding and obstructed defecation. Therapeutic experience in children with SRUS, is limited, with variable treatment protocols and outcomes. Randomized controlled studies documenting long-term follow-up to establish optimum treatment in children are required.

References