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Case series with literature review: Surgical approach to megarectum and/or megasigmoid in children with unremitting constipation



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ABSTRACT

Background: The role of surgery in treating children with functional constipation (FC) is controversial, because of the efficacy of bowel management programs. This case series is comprised of failures: 43 children, spanning 25 years' practice, who had megarectosigmoid (MRS) and unremitting constipation.

Purpose: To determine whether these children were helped by surgery, and to contribute to formulating a standard of care for children with megarectum (MR) and/or redundancy of the sigmoid colon (MS) who fail medical management.

Method: We describe our selection criteria and the procedures we utilized – mucosal proctectomy and endorectal pull-through (MP) or sigmoidectomy (SE) with colorectal anastomosis at the peritoneal reflection. The internet (social media) allowed us to contact most of these patients and obtain extremely long follow-up data.

Results: 30/43 patients had MP and 13/43 had SE. Follow-up was obtained in 83% MP and 70% SE patients. 60% of MP and 78% of SE patients reported regular evacuations and no soiling. 20% MP patients had occasional urgency or soiling or episodic constipation. 12% MP and 22% SE patients required antegrade continence enemas (ACE) or scheduled cathartics and/or stool softeners. 4% MP had no appreciable benefit, frequent loose stools and soiling, presumably from encopresis.

Conclusion: MR is characterized by diminished sensation, poor compliance and defective contractility. Patients with MR do better with MP, which effectively removes the entire rectum versus SE, where normal caliber colon is anastomosed to MR at the peritoneal reflection; furthermore, MP reliably preserves continence; whereas total proctectomy (trans-anal or trans-abdominal) may cause incontinence.

1. Introduction

Functional Constipation (FC) is a disorder of homeostasis. With less frequent defecation, stool accumulates and stagnates. Increased water absorption results in bulky, hard stools; and pain during defecation promotes stool retention. A vicious cycle ensues. FC patients have symptoms of intestinal obstruction: bloating, cramps, and diminished appetite. Ultimately, their health and wellbeing suffer.

1.1. Table 1 Characteristics of children with severe constipation [1]

Bowel management programs usually are effective in treating FC [2]; however, not all patients tolerate laxatives, especially the large doses that are prescribed; stool softeners may exacerbate soiling [3];

and many children simply refuse enemas [4].

FC is a heterogeneous disorder, as physiologic investigations have demonstrated. More precise understanding of pathogenesis promotes therapeutic efficacy. Colonic manometry and radionuclear transit distinguish "slow colonic transit" (SCT) from "fecal retentive constipation" (FRC), which is usually associated with MR [5]. Anal sphincter resting pressure usually is normal in MR [6] but increased in anal sphincter achalasia and dyssynergia. The anorectal inhibitory reflex is present in MR but absent in patients with Hirschsprung's Disease.

In our estimation, the contrast enema remains the most valuable tool in evaluating children with unremitting constipation. Static radiographs define anatomy; and post-evacuation films document adequacy of rectal emptying. Defecography is a more accurate study but has limited usefulness in children.

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 Table 1

 Characteristics of children with severe constipation.

Children with Functional Constipation have two or more of these characteristics:

1.

Two or fewer defecations/week

2.

One or more episodes of fecal incontinence/week

2

Retentive posturing or volitional stool retention

Painful or hard bowel movements

5.

Rectal stool bolus palpable by abdominal exam

6.

Large diameter stools obstructing the commode

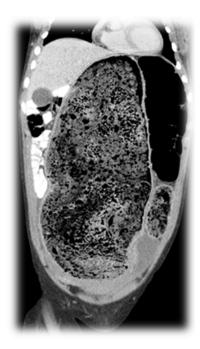


Fig. 1. Megarectosigmoid simulates a gravid uterus in this adolescent girl.

MR has distinguishing radiologic features, other than distension (Fig. 1). The normal rectum has an oval shape, and it is located in the pelvis. MR is conical in shape and extends above the pelvic brim. The diameter of MR is maximal at the sacral promontory; and usually, there is an abrupt transition between the hugely dilated rectum and normal caliber colon [7].

1.2. Fig. 1 Adolescent girl whose megarectum simulates a gravid uterus!

MR was first described in association with anorectal malformations [8,9]. Later, it was recognized in otherwise normal children with unremitting constipation. Various terms have been used to describe MR: atonic baggy rectum, terminal fecal reservoir, rectal ectasia and inertia [10]. It may present during infancy or later (Figs. 2 and 3). The postulated causal sequence is as follows:

- 1. An enteric nerve or muscle abnormality
- 2. Impaired peristalsis resulting in distension
- 3. Excess stretch weakening muscular contraction
- 4. Dysmotility leading to incomplete evacuation and stool retention
- 5. Fecal impaction and ultimately encopresis (overflow incontinence)



Fig. 2. Megarectum in an infant.



Fig. 3. Megarectum in an adolescent.

1.3. Figs. 2 and 3 Infant and adolescent with megarectum

Continence requires a compliant rectum with effective peristalsis and an anal sphincter that contracts, generating an adequate resting pressure. In patients with ARM and MR, it may be difficult to distinguish true incontinence (inadequate sphincter) from pseudoincontinence (overflow). Recovery of continence following proctectomy may be predicted by assessment of anal sphincter function with manometry and anal sphincter musculature by endosonography [6].

Manometry in patients with MR requires inflation of the balloon to supernormal volumes. A normal reading is encouraging; these patients may respond to bowel management programs. If the rectum is stiff and noncompliant, contractility is impaired, proprioception (perception of distension) is diminished, and the anorectal inhibitory reflex is attenuated. Medical intervention is unlikely to be effective in such patients.

Patients with MR are outliers in the FC spectrum. Typically, these children have always had difficulty moving their bowels. They were never "toilet trained"; they are incontinent. MR's huge capacity predisposes to fecal impactions, painful defecation, and encopresis. These children usually have seen multiple practitioners; their parents are frustrated, at their wits' end; whereas the child may present a façade of *insouciance* - "la bell indifference." Self-control is a developmental milestone; these children cannot even control their bowel movements, a situation made all the more devastating because it is misunderstood. People assume that they are incontinent by choice and that they choose not to exercise self-control. As an example of the suffering caused by this disorder, one young man was required to sit in a particular chair. He could not choose where he sat, because he was considered unclean, like a leper.

MR occurs infrequently, comprising only 11% of patients referred to tertiary colorectal center for treatment of refractory constipation [3]. This low incidence may account for the lag in acceptance of MR as an

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