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Persistent rectal prolapse in children: sclerotherapy and surgical management

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Abstract Persistent rectal prolapse is an uncommon but distressing condition in children. Significant controversy exists regarding its surgical management. The aim of this study was to identify a successful management strategy for persistent rectal prolapse in the paediatric population. Records of all children with rectal prolapse treated surgically at Birmingham Children's Hospital between 1995 and 2003 were retrospectively reviewed. Demographic data, clinical presentation, investigations, treatment modality, complications, and outcome were recorded. Inclusion criteria for the study were failure of conservative management leading to operative treatment. An exclusion criterion was cystic fibrosis. A total of 24 patients with persistent rectal prolapse were identified. Two children with cystic fibrosis were excluded from the analysis. Children below the age of 5 years, group I ($n=17$), were successfully managed by submucous hypertonic saline injections. Eighty-three percent (14/17) were cured by injection sclerotherapy in this group, 12/14 (71%) requiring one injection and 2/14 requiring a second injection. In the three (17.6%) children in group I in whom sclerotherapy failed, cow's milk protein (CMP) allergy was identified as the causative factor. Children older than 5, group II ($n=5$), either had behavioural problems ($n=3$) or were autistic ($n=2$). This group of children with adult-type, full-thickness rectal prolapse were found to be refractory to initial attempts of injection sclerotherapy. All five children were successfully managed with surgical correction. We conclude that rectal submucous hypertonic saline injections are highly effective for managing early-onset idiopathic childhood rectal prolapse. CMP allergy should be considered in young children with recurrent rectal

prolapse. We recommend early definitive corrective surgery in older children with persistent rectal prolapse, as they do not respond to conservative measures or injection sclerotherapy.

Keywords Rectal prolapse · Children · Hypertonic saline injection

Introduction

Persistent rectal prolapse is a well-recognised but relatively uncommon condition in children. In western countries aetiology is usually related to excessive straining, constipation, and functional defaecation disorder rather than diarrhoea, dysentery, and malnutrition-related rectal prolapse, as seen in third-world countries [1]. The prolapse is usually precipitated by the increased intraabdominal pressure related to an excessive straining effort at defaecation and may be associated with some anatomical configuration of the rectum and pelvis or loosely attached mucosa to the underlying muscularis of the rectal wall [2].

There are two types of paediatric rectal prolapse. One is the intermittent self-limiting variety, which is more common, less pronounced, and responsive to conservative measures. The other variety recurs with every defaecation or straining and may require manual reduction, causing significant anxiety and distress both to the child and to the parents. Although conservative measures can be successful in some cases, surgical intervention is required for recurrent and distressing rectal prolapse. Many surgical procedures to contain rectal prolapse in children are advocated but without any coherent evidence-based approach. Surgical procedures ranging from the less invasive injection sclerotherapy [3] to more aggressive surgery, including abdominal posterior rectopexy [4], abdominal or perineal bowel resection [5], transanal suture rectosacropexy [6], and posterior sagittal procedures [7] have been described. The large

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number of described procedures lacks consensus and evidence to reflect their success in all cases of rectal prolapse in children. The surgical principles for managing rectal prolapse in adults are usually directed either at narrowing the anal orifice, suspending the rectum, shortening the redundant rectum, or restoring the enlarged pelvic floor and levator hiatus. The aim of this study was to review our experience with the treatment of persistent rectal prolapse in children and to formulate a management strategy based on outcome.

Materials and methods

Records of all children with rectal prolapse treated surgically at the Birmingham Children's Hospital between 1995 and 2003 were retrospectively reviewed. Twenty-four patients were identified.

Data collected included age, gender, and clinical presentation, including number of visits to the emergency department, past medical history, clinical examination, investigations, treatment, complications, and outcome.

Investigations included a sweat test in all patients to exclude cystic fibrosis and a stool examination for parasites. Children with rectal prolapse associated with loose motions were investigated for infective causes, coeliac disease, and fat malabsorption. All children were initially managed conservatively for constipation and straining, using stool softeners, laxatives, a high-fibre diet, and adequate fluid intake. Children were included in the study if they had persistent and recurrent rectal prolapse for several months despite medical management or that required repeated reductions or repeated visits to the emergency department. The data on successful conservative management of rectal prolapse were unavailable because these children were not referred for the surgical management.

Patients were divided into two groups according to age at the time of presentation. Group I comprised children younger than 5 years, and group II comprised older children. The main presenting symptom was a mass coming out of the rectum. Injection sclerotherapy was the first line of surgical treatment in all patients. They underwent sigmoidoscopy and examination under anaesthetic to rule out any local cause, such as a rectal polyp, prior to submucous hypertonic saline sclerotherapy.

Technique of submucous injection sclerotherapy

The operation was performed under general anaesthesia with a caudal block for postoperative analgesia. The children were placed in lithotomy position. The buttocks and perianal area were prepared with povidone-iodine. After digital examination of the rectum, sigmoidoscopy was performed. Hypertonic saline (15%) was injected in the submucous plane in all four quadrants of the rectum 1–2 cm above the dentate line with a 21-gauge needle. A

maximum of 10 ml (2–2.5 ml in each quadrant) was injected. The procedure was carried out as a day case. Children were discharged home to continue with their stool softeners, diet high in fibre and fluids, and oral analgesics.

All children were followed up in outpatient clinic 4–6 weeks after the sclerotherapy and operative procedure. Subsequent follow-up was arranged after 3–6 months depending on symptoms.

Results

A total of 24 patients were identified. Two patients with inappropriate straining and loose motions were diagnosed to have cystic fibrosis on sweat test and were managed with pancreatic enzyme replacement. They were excluded from the study.

Group I comprised 17 children aged 2–4.5 years (median 2.5 years). Thirteen children had constipation with straining, whereas loose motion was the associated symptom in two children. Six children attended the emergency department more than once during the conservative management period with distress and persistent rectal prolapse with as many as six visits in one child. The length of prolapsed bowel varied greatly from child to child.

In group I ($n=17$), all children were successfully managed with submucous hypertonic saline injections. Seventy-one percent (12/17) were managed successfully with a single injection, whereas the success rate after two injections was 83% (14/17). In the remaining three (17%) children, cow's milk protein (CMP) intolerance was identified as the cause of recurrence because after excluding CMP from the diet, no recurrence was observed. The recurrences of rectal prolapse in these children were immediate after hypertonic saline injections, and prolapse was much more debilitating and occurred after every defaecation. The exclusion of CMP in their diets resulted in cure with no recurrences. The lack of recurrences is unlikely to be related to the effect of injection or to a spontaneous cure; in one child, the parent restarted CMP in the diet, which resulted in recurrence. There were no complications following 15% hypertonic saline injections. Asymptomatic patients were discharged from follow-up after 1 year.

Children in group II ($n=5$) were aged 5–13 years (median 12 years). All children in this group presented with full-thickness adult-type rectal prolapse. Two had autism, and three had significant functional defaecation disorder with behavioural problems. Four children in this group had one or more unsuccessful attempts of perirectal hypertonic saline injection (range: one to three) to treat rectal prolapse. Surgical correction in group II included Thiersch anorectal encircling with a nonabsorbable suture (one), Ivalon sponge implant (one), and proctosigmoidectomy (three). The selection of surgery type in this group depended on the identified anatomical defect. There were no reported complica-

tions or recurrences after the surgical correction in this group in follow-up.

Discussion

Rectal prolapse is associated with a variety of conditions that cause either increased or prolonged straining associated with idiopathic constipation, Hirschsprung's disease, and cystic fibrosis [8–10]. It can also occur with dehydration and malnutrition associated with acute or chronic diarrhoea and parasitic diseases [11–14]. Pelvic floor weakness as seen in adults is usually not the factor in children. Functional defaecation disorders and prolonged straining associated with constipation are noted to be frequent causes of prolapse in older children in western countries. Patients should be investigated to identify predisposing conditions such as cystic fibrosis, parasitic infestation, and coeliac disease. Treatment of the underlying cause resolves the prolapse in most instances. However, prolonged persistent rectal prolapse causes significant distress to the patient and parents, warranting surgical intervention [7].

The pathophysiology of rectal prolapse is relevant because it has implications on treatment, as shown in this study. Prolapse probably begins at the mucocutaneous junction because of a shallow mucocutaneous sulcus. This is characteristic of the rectal prolapse in infants regardless of the length of the prolapse [15]. In older children and adults, the prolapse is full-thickness, with invagination beginning at the rectosigmoid level [16]. The classic abnormalities found in these patients—wide deep pelviperitoneal pouch, unsupported redundant rectum with long mesorectum, weak pelvic floor and anal sphincter—are probably effects rather than causes [4].

Injection of hypertonic saline is the most commonly used minimally invasive technique for rectal prolapse. Many different agents have been described as successful [17–19]. We used submucosal injection of 15% hypertonic saline with good success and no complications. Submucosal injection helps to avoid complications such as ischiorectal abscess and perirectal inflammation [20]. In group I, 12/17 (71%) patients were cured after one injection, and 14/17 (83%) after two injections. Kay and Zachary [17] reported a success rate of 78% and 94%, whereas Dutta and Das [20] reported a success rate of 83% and 97% using one and two injections of 30% saline. Complications, including perirectal inflammation, ischiorectal abscess, and necrosis of rectal mucosa, have been reported using 30% saline [3]. Submucosal injection of reduced concentration (15%) avoided this morbidity with a good success rate in our patients.

CMP intolerance was the precipitating cause of recurrence in three children in group I. Cow's milk was empirically stopped in children with recurrences after the third treatment of hypertonic saline injections, resolving the symptoms and the prolapse. Although the underlying mechanism is still unclear, CMP intolerance is a well-

established cause of chronic constipation [21, 22]. Resolution of symptoms is unlikely to be coincidental or due to a spontaneous cure, as the effect was immediate. Also, reinstatement of CMP in one child resulted in recurrence of the rectal prolapse. This is the first report implicating CMP intolerance as an underlying cause of recurrent rectal prolapse in children.

The older children in group II presented with adult-variety full-thickness rectal prolapse. Similar findings were reported by other series [23]. These children do not respond to submucosal or perirectal sclerosing injection therapy, as shown in our series and other authors' series [23]. Analysis of the age of patients successfully treated for rectal prolapse with various surgical techniques other than hypertonic saline shows that most of these children were older than 4 years [6, 23]. These data further substantiate our clinical age grouping and the fact that older children would benefit from aggressive surgical therapy. Indiscriminate surgical correction of rectal prolapse in children of all ages is not indicated, as hypertonic saline (15%) is safe, extremely effective, and less invasive.

Two older children in our group II were autistic. Full-thickness rectal prolapse occurs more frequently in autistic children than in the general population [24]. Rather than being a congenital condition, rectal prolapse in these children is more of a developmental condition that manifests over time, hence the delayed presentation [25]. It is clear that these children do not seem to respond to efforts aimed at minimising straining with defaecation and therefore are refractory to medical management and hypertonic saline injections. A definitive surgical intervention was thus deemed inevitable. We agree with others that this group of children would benefit from early definitive corrective surgery to contain rectal prolapse rather than giving them a trial of conservative surgery.

Three children in group II had significant functional defaecation disorder. Rectal prolapse has been reported to occur in children with behavioural problems and mental retardation [26, 27]. Prolapse is usually due to straining at defaecation against a closed sphincter mechanism. Such straining may be obsessive on the part of patients with psychosocial problems. These children had multiple trials with medical management consisting of stool softeners, dietary manipulation, colonic evacuation, bowel training, and behavioural management, without any success. The length of medical management was unacceptably long and distressing for the patients and parents; therefore, an earlier surgical option should be considered. These children in our study were found to be refractory to injection sclerotherapy, and an eventual definitive surgical correction was considered. The surgery should be aimed at correcting the identified disorder. One child was treated with Thiersch anorectal encircling suture as the first instance, whereas two children had proctosigmoidectomy with recurrence-free results. Our series is too small to recommend or formulate any specific type of surgical strategy for this group of children. However, we do suggest that therapy should be

aimed at the anatomical defect rather than chosen because of familiarity of the surgical procedure. Random selection of the surgical procedure in these children may be related to the failure of surgery and to recurrence.

Our findings and results substantiate that rectal prolapse in children in the younger age group (< 5 years) begins with mucosa at the mucocutaneous junction and proceeds in some cases to full thickness of the bowel. The aim of surgical intervention in these cases should therefore be creation of fibrous adhesion between the mucosa and the muscle wall of the rectum—hence the reported success of submucous sclerosant injections. The late onset full-thickness adult-type rectal prolapse did not respond to sclerotherapy in our study, and such patients would benefit from more aggressive surgery at an earlier stage. Choice of surgery varies from patient to patient, as suggested by investigations and the surgeon's familiarity with a particular procedure to correct the pathology.

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