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A systematic review of management options in pediatric rectal prolapse☆

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ABSTRACT

Purpose: Rectal prolapse is a relatively common condition in infants and young children with a multifactorial etiology. Despite its prevalence, there remains clinical equipoise with respect to secondary treatment in pediatric surgery literature. We conducted a systematic review to evaluate methods of secondary treatment currently used to treat rectal prolapse in children.
Methods: We searched Pubmed, Medline, and Scopus with the terms "rectal prolapse" and "children" for papers published from 1990 to April 2017. Papers satisfying strict criteria were analyzed for patient demographics, intervention, efficacy, and complications. Procedures were grouped by like type. Pooled success rates were calculated.
Results: Twenty-seven studies documenting 907 patients were included. Injection sclerotherapy had an overall initial success rate of 79.5%. Ethyl alcohol seemed the best sclerosing agent due to a high first-injection success rate, low complication rate, and ready accessibility. Several perineal repairs were found, with operative success rates ranging from 60.8%–100%. Laparoscopic rectopexy with mesh was the most commonly reported transabdominal procedure and had an overall success rate of 96.1%. Postoperative complications from all procedures were comparable.
Conclusion: Though many secondary treatment options have been reported for rectal prolapse, sclerotherapy and laparoscopic rectopexy predominate in contemporary literature and appear to have high success and low complication rates.
Level of Evidence: IV.

Contents

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Rectal prolapse continues to be a significant issue for children due to anatomic features of the pediatric pelvis and early toileting habits. Weak pelvic levator musculature and loose attachment of the rectal mucosa to the muscularis predispose young children to the condition, and straining during stooling or prolonged periods of sitting on the toilet exploit these normal anatomic features to result in protrusion of the rectal mucosa or full-thickness rectal wall through the anal canal [1,2]. Though usually painless, rectal prolapse is a distressing condition that is often brought to quick medical attention. The primary treatment of rectal prolapse is nonoperative, through encouraging a high fiber diet, administering stool softeners, minimizing straining during defecation, and discouraging prolonged toilet sitting. The success rate of primary treatment has been reported to be 28–50% [3–5]. Secondary treatment consists of surgical procedures designed to prevent the rectum from prolapsing. Over 100 surgical procedures have been documented in the literature to treat rectal prolapse, indicating a lack of consensus from the surgical community as to the most efficacious treatment of this condition.

In order to ease this clinical quandary, we sought to identify the most common procedures used to treat pediatric rectal prolapse in primary, idiopathic cases; quantify and review their success rates; and describe post-procedure complications.

1. Methods

We performed a systematic review of the literature to identify the various methods of secondary treatment of pediatric rectal prolapse. This systematic review follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [6].

1.1. Selection criteria

Inclusion criteria for analysis in our review were a primary source report of: (a) specific procedural treatment for rectal prolapse in patients 18 years of age or younger and (b) post-treatment outcomes and complications. Papers needed to include at least five patients to be considered.

We excluded: (a) papers discussing multiple interventions with no breakdown of post-treatment outcomes by specific procedure; (b) papers in which adult outcomes were unable to be separated from pediatric outcomes; (c) papers primarily evaluating rectal prolapse secondary to infectious disease, neurological malformation such as myelomeningocele, psychiatric disorder, or unique circumstances; (d) papers without discussion of follow-up; and (e) papers whose full text was unavailable. References in the articles chosen for study analysis were examined for additional papers to be included.

1.2. Search strategy

We performed a comprehensive literature search using multiple databases. The databases included were Medline, PubMed, and Scopus. The search terms used were “rectal prolapse” and “children.” Results were limited to the English language and human species and restricted by publication date to 1990 through April of 2017. We additionally reviewed the listed references in the papers meeting inclusion and exclusion criteria to identify any other studies suitable for our review.

1.3. Data abstraction and quality assessment

Variables collected were study type, study location, study period, number of patients, patient age and gender, type of intervention, specific intervention protocols, follow-up period, recurrences, postoperative outcomes (constipation, continence, ongoing use of medications, and cosmesis), and postoperative complications. Papers were separated into those discussing sclerotherapy and those discussing operative intervention. Sclerotherapy papers were categorized based on sclerosing agent. Operative interventions were categorized into perineal repairs and transabdominal repairs and grouped within those categories by like procedures. Bias in our results was assessed at the outcomes level, considering the global impact of the studies we assessed.

1.4. Statistical methods

Pooled efficacy rates, defined as the number of patients successfully treated without recurrence divided by the total number of patients treated, for each type of procedure were calculated and reviewed. In order to simplify categories, procedures were grouped by primary intervention. For example, several papers reported modifications of anal suture cerclage, with different puncture sites or types of suture; these were all grouped together. Since most studies were case series designs with no control/comparison group, no meta-analysis was conducted. Instead, the combined results were reviewed for a pattern in efficacy.

2. Results

2.1. Cohort assembly

Our database search yielded 248 publications related to rectal prolapse (Fig. 1). Of these, 58 publications satisfied both inclusion criteria. No additional publications were identified through review of these articles' references. Thirty-one papers were excluded based on exclusion criteria. Twenty-seven publications documenting the treatment of 907 patients remained for analysis in this review.

2.2. Demographics

Publications included in our review span ten countries in North America, Europe, Asia, and Africa. Patient ages ranged from infancy through adolescence, with one patient, a 19-year-old, in one study

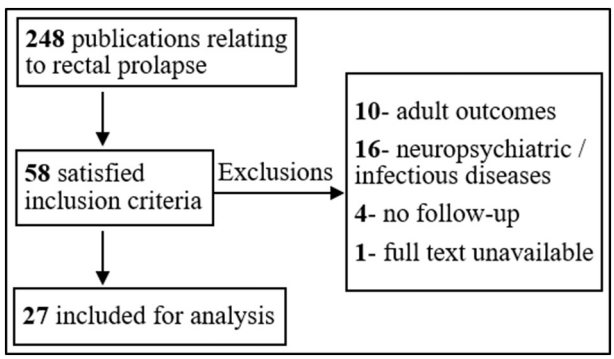

Fig. 1. Literature search flowchart.

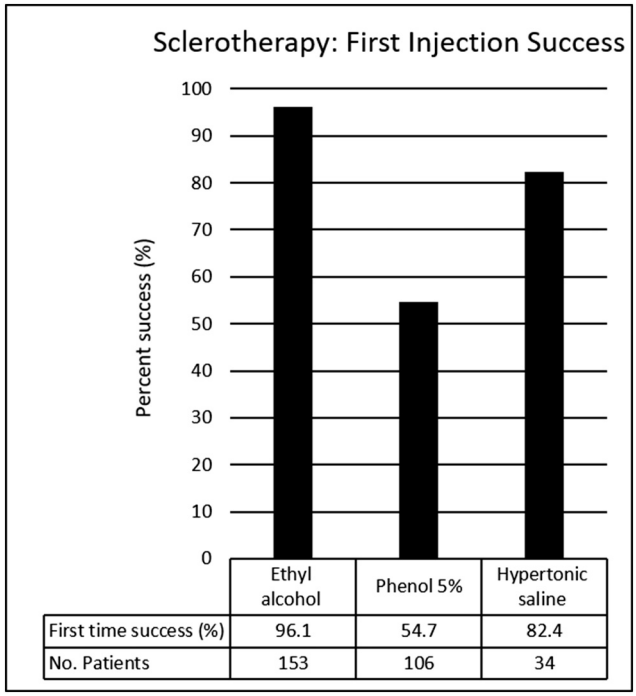

Fig. 2. Number of patients undergoing sclerotherapy and first-time injection success rates, by sclerosing agent.

being over age 18 years at the time of treatment. Gender breakdown was given for 476 of the 907 patients; 60.3% of these patients were male.

2.3. Interventions

We found eight studies describing injection sclerotherapy treatment of 305 patients with three different sclerosing agents. The sclerosing agents used were: 96% ethyl alcohol [7], 5% phenol [8–11], and hypertonic saline [12–14].

We found 22 studies discussing operative management of 602 patients with at least 17 operative procedures. These operative interventions can be broadly classified by approach: perineal or transabdominal. The most common perineal repairs included several variations on intestinal pexy, mucosal plication [15,16], and anal suture cerclage [8,13,14,17,18]. Perineal rectopexy strategies included posterior sagittal rectopexy [13,19,20], transanal rectosacropexy (Ekehorn's procedure) [21,22], closed rectosacropexy [23], and retrorectal gauze rectopexy (the Lockhart-Mummery procedure) [24]. The most common transabdominal repair performed was also rectopexy: open rectopexy with mesh [13,24], laparoscopic rectopexy with mesh [25–29], or laparoscopic rectopexy with suture [8,13,27,28,30,31].

2.4. Treatment efficacy

First injection success rates across all three sclerosing agents ranged from 54.7% to 96.1% (Fig. 2). Ethyl alcohol appeared to have the highest success rate in the first injection at 96.1%. Hypertonic saline appeared to be less efficacious with an 82.4% first injection success rate, and 5% phenol appeared to be the least efficacious of these three agents with a 54.7% first injection cure rate.

First-time success rates of perineal procedures can be seen in Fig. 3. Transanal rectosacropexy, closed rectosacropexy, retrorectal gauze rectopexy, and mucosal plication all had first-time success rates over 98%. Posterior rectopexy and suture cerclage (the Thiersch stitch) appeared to have lower first-time success rates, at 77.1% and 60.8%, respectively.

First-time success rates of transabdominal procedures can be seen in Fig. 4. Of the three main types of intestinal pexy, open rectopexy with mesh appeared to have the highest first-time success rate at 98.4%. Laparoscopic rectopexy with mesh had a 96.1% first-time success rate, and laparoscopic rectopexy with suture had an 81.4% first-time success rate. Transabdominal intestinal resection (either open or laparoscopic) [14,27] and posterior rectopexy with omental pedicle [32] also had high success rates, but we found documentation of fewer than 20 patients total undergoing these procedures.

2.5. Complications of sclerotherapy

Complications reported from sclerotherapy studies were rare. Temporary limping lasting 2–3 days was reported in four of 165 patients treated with ethyl alcohol sclerotherapy by Bahador [7]. This was felt to be due to infiltration of the alcohol outside the rectal wall at the level of the sciatic nerve. No infectious complications were reported in any of the studies using ethyl alcohol, phenol, or hypertonic saline as sclerosing agents.

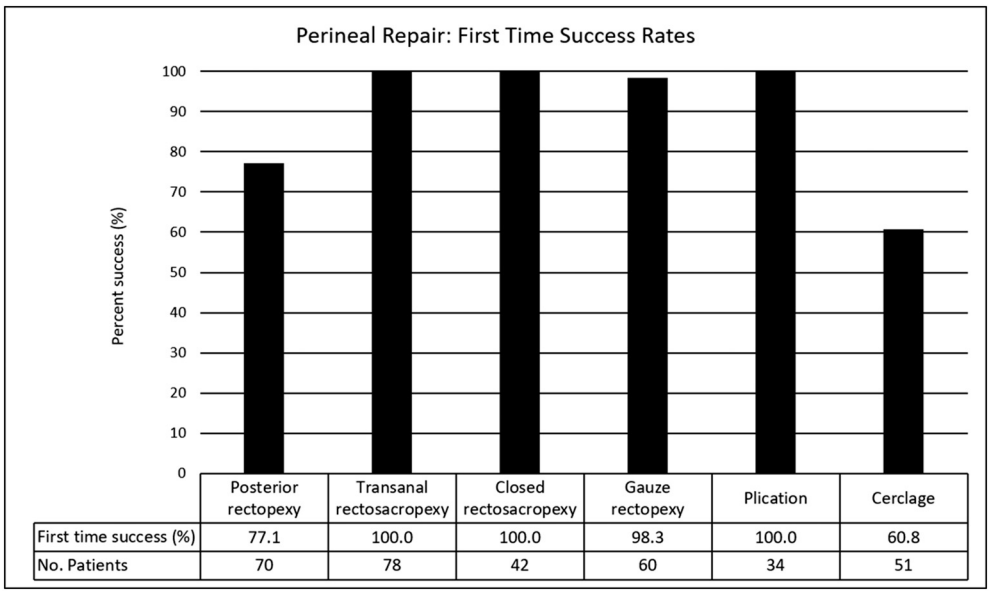

Fig. 3. Number of patients and first-time success rates of perineal repair procedures, by type of repair.

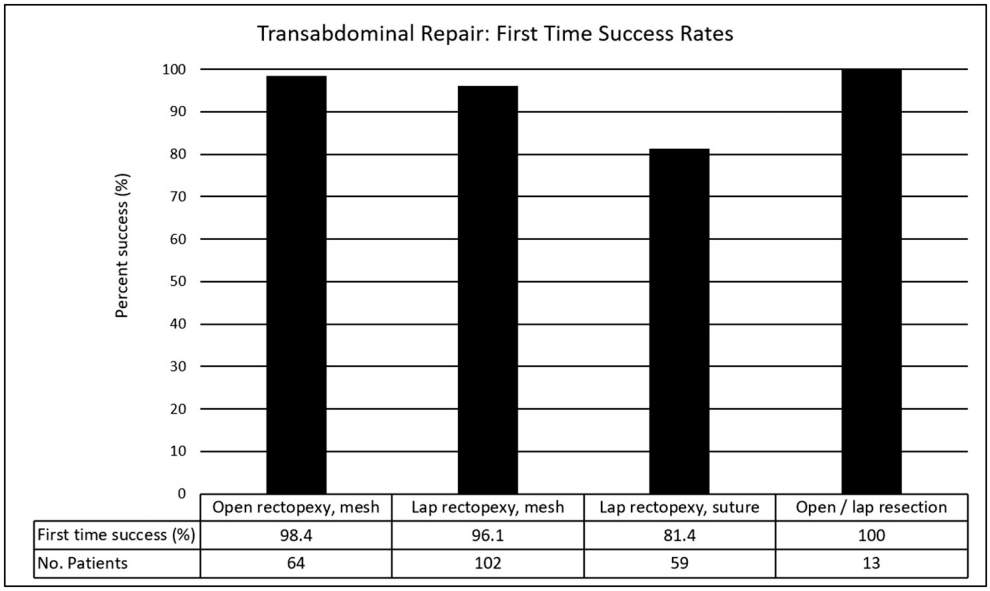

Fig. 4. Number of patients and first-time success rates of transabdominal repair procedures, by type of repair.

2.6. Complications of operative interventions

Nazem and colleagues compared open transabdominal rectopexy and perineal retrorectal gauze rectopexy (Lockhart-Mummery procedure) in the only randomized controlled trial we found [24]. The authors found no statistically significant difference in complication rates between the two procedures, though perineal retrorectal gauze rectopexy trended towards a lower complication rate.

Complication rates of the most commonly-performed perineal procedures can be seen in Table 1. The most common complication overall was superficial wound infection, which was to be expected following transanal rectosacrpoexy, as Sander points out [21]. These infections presented with mild fever, erythema around the suture sites, and sometimes mild pus evacuation from the suture holes. Other perineal procedures, including transanal mucosal sleeve resection (modified Delorme procedure), anal stretching, banding, and the Ivalon sponge implant cited no complications but were rarely reported [8,14,33]. Overall, among the main procedures reported, posterior rectopexy and retrorectal gauze rectopexy appeared to have the lowest rates of postoperative complications.

Table 1
Complications of perineal repairs. No complications were reported for the other perineal repair types.

Complication	Rate (%)
Posterior rectopexy	**7.2**
Extrusion of sutures	*2.9*
Wound infection	*2.9*
Unrelated death	*1.4*
Transanal rectosacropexy	**64.1**
Localized infection	*64.1*
Gauze rectopexy	**3.4**
Wound infection	*1.7*
Bleeding	*1.7*
Suture cerclage	**20.5**
Unrelated death	*1.7*
Suture cut through	*3.4*
Wound infection	*1.7*
Perianal abscess	*1.7*
Rectal bleeding	*5.2*
Fistula-in-ano	*1.7*
Rectal narrowing	*1.7*
Re-operation	*3.4*

The complication rates reported from the transabdominal procedure can be seen in Table 2. One of the 13 patients undergoing intestinal resection developed a temporary colocutaneous fistula which responded to conservative treatment [27]. Surgical intraoperative complications were reported in none of the patients undergoing laparoscopic rectopexy across nine studies [8,13,25–31], though Awad did report one laparoscopic suture rectopexy needing conversion to an open procedure due to equipment malfunction and one patient who developed a port site hernia [30].

2.7. Secondary outcomes

The main secondary outcomes reported were continence, constipation, ongoing medication usage, and cosmetics. These outcomes were inconsistently reported.

In Bahador's study, 25 of 153 patients (15.2%) exhibited fecal soiling for a few days following injection sclerotherapy with ethyl alcohol [7]. Two of four patients (50.0%) were reported by Sasaki to have resolved incontinence after sclerotherapy with phenol [11].

Constipation and ongoing medication use were reported in several studies. In laparoscopic rectopexy, constipation was reported in 1.9–7.1% of patients [26,27,29]. Shalaby routinely prescribed lactulose to prevent constipation for 1 month postoperatively [29], and Potter reported use of stool softeners in seven of 19 patients (36.8%), laxatives in six of 19 patients (31.2%), and antegrade enema in two of 19 patients (10.5%) [31].

Table 2
Complications of transabdominal repairs. No complications were reported for the other transabdominal repair types.

Complication	Rate (%)
Open rectopexy, mesh	**15.7**
Wound infection	*6.3*
Bleeding	*4.7*
Adhesions	*4.7*
Laparoscopic rectopexy, suture	**5.1**
Conversion to open	*1.7*
Supplementation w/ suture cerclage	*1.7*
Port site hernia	*1.7*
Intestinal resection	**7.7**
Temporary coloenteric fistula	*7.7*

Little was reported on cosmetic outcomes. Two studies utilizing laparoscopic techniques (rectopexy and resection) reported "excellent" cosmetic results [27,29]. Elmalik claimed "satisfactory" cosmetic results for all five patients undergoing posterior sagittal rectopexy [32], and Lasheen noted that the postanal stab incisions healed well in all patients after closed rectosacropexy [23].

3. Discussion

Our systematic review of secondary management options in pediatric rectal prolapse identified 27 publications documenting the treatment of over 900 patients. One-third of these patients underwent sclerotherapy with one of three sclerosing agents, while the remaining underwent operative intervention. At least 17 operative interventions were identified.

Rectal prolapse is more likely to be self-limiting in children under 4 years of age, with cases presenting later in childhood often found secondary to predisposing conditions such as diarrheal illnesses, cystic fibrosis, and malnutrition. After ruling out underlying conditions, nonoperative management should therefore be the first course of action in most cases. Consuming a high-fiber diet, taking stool softeners and laxatives as needed, providing children with a step stool in front of a regular toilet, and limiting time spent on the toilet can all help to maintain a regular bowel schedule and prevent straining [1].

The duration of nonoperative management should be tailored to the individual, with the decision to pursue procedural intervention dependent on symptom severity, frequency of prolapse, patient age, and patient and parental distress. Frequent, bothersome prolapse should be repaired in a timely fashion. Several authors have suggested pursuing nonoperative management strategies for up to 1 year in less severe or less frequent cases [8,31]. It has also been pointed out that delaying repair of less bothersome prolapse until after 4 years of age may be beneficial, as earlier surgical correction has been associated with recurrences and difficult reduction [8].

Injection sclerotherapy is the least invasive secondary treatment method. A sclerosing agent is injected just above the dentate line into the submucosal space creating an inflammatory reaction that causes adherence of the elements of the anorectum. Of the three sclerosing agents identified in our review (96% ethyl alcohol, 5% phenol, and hypertonic saline), ethyl alcohol had the highest first-injection cure rate at 96.1%. Complications after injection sclerotherapy were typically benign and short-lived.

Surgical interventions for rectal prolapse are generally aimed at reducing the immediate protrusion and restoring normal physiology by fixing the rectum, narrowing the anal orifice, resecting redundant intestine, and/or strengthening the pelvic floor. The specific goals of each procedure are different, and therefore the choice of operation must be tailored to each individual patient. For example, the goal of anal cerclage (Thiersch stitch) is simply to reduce the circumference of the anal orifice, thereby decreasing the possibility of recurrent prolapse. The transanal mucosectomy and muscularis plication (Delorme procedure) removes rectal redundancy by essentially decreasing the length of the rectal mucosa. Rectopexy aims to prevent prolapse by fixing the rectum to the sacral prominence. Several procedures, including transanal rectopexy, closed rectosacropexy, retrorectal gauze rectopexy (Lockhart-Mummary procedure), and transabdominal rectopexies attempt to achieve this goal via different approaches. Patients with a redundant rectum and/or sigmoid colon may need to have intestinal resection, which can be performed via a transanal approach (Altemeier procedure) or an open or laparoscopic transabdominal approach.

In our review, the efficacy of several perineal rectopexy procedures was found to be high, with transanal rectosacropexy, closed rectosacropexy, and retrorectal gauze rectopexy each having a reported success rate of greater than 98%. Transanal rectosacropexy (Ekehorn's rectosacropexy) involves passing a mattress suture from inside to outside through the rectal ampulla, creating a "U" loop of suture at the level of the sacrococcygeal junction. A piece of dry gauze is placed inside the "U" loop, and the suture is tied externally and left in place for 1–2 weeks. Closed rectosacropexy entails passing several sutures through 3–5 perineal stab incisions. One arm of each suture passes from the rectal vault through the presacral fascia, and the other arm passes from the rectal vault in a submucosal plane. The Lockhart-Mummery retrorectal gauze rectopexy is a type of open sclerosing procedure no longer routinely performed in which the retrorectal space is developed from the perineum and packed with gauze. Suture cerclage is another commonly-reported perineal procedure that we found to have a lower overall efficacy rate at 60.8%. A concern in performing this procedure is overtightening the suture, though this risk can be reduced by tying over an assistant's finger or a Hegar dilator [1].

With the advancement of laparoscopy in recent decades, laparoscopic rectopexy has largely replaced many perineal procedures. In our review, laparoscopic rectopexy was found to be efficacious and safe, with only infrequent and mild complications, such as conversion to an open procedure and port site hernia, that would not be unexpected with other laparoscopic procedures. Laparoscopic techniques utilizing either mesh or suture rectopexy have been reported. In a prospective database review, Randall found a tendency towards reduced prolapse recurrence with mesh rectopexy compared to suture rectopexy [28]. While findings in our review agree with this success rate stratification (96.1% success for mesh rectopexy vs 81.4% for suture rectopexy) and show no complications related to mesh use in laparoscopic rectopexy, we would caution the reader of the potential for high morbidity – including mesh erosion, infection, sepsis, and the need for reoperation – with use of synthetic mesh [2,28]. Literature reporting on the use of mesh in the repair of pelvic organ prolapse cites a 4.2% rate of mesh complications in adults undergoing sacrocolpopexy, a procedure analogous to rectopexy, and a 2010 systematic review reported a synthetic mesh erosion rate of up to 12% [34,35]. In line with this evidence, a 2014 consensus statement advocates for the use of biologic over synthetic material when mesh is to be used in young or adolescent patients [36].

3.1. Approach to a patient with rectal prolapse

Evaluation of a child with rectal prolapse begins with a thorough history and physical exam. Cystic fibrosis and parasitic gastrointestinal infections should be ruled out. Duration and frequency of episodes, length of protrusion, and impact on stress levels and quality of life for the parents and patient should be assessed. If the prolapse is not reproducible in the office by having the child strain on a commode, a simple cell phone picture from the parents is usually enough to make the diagnosis. Nonoperative management as discussed above should be the first course of treatment. Indications for procedural intervention include mucosal ulceration with bleeding, frequent prolapse, requirement for manual reduction, frequent hospital visits with irreducible prolapse, failure of nonoperative management, and reluctance of patient or parents to continue with nonoperative management [21].

Proper selection of the appropriate procedure for a given patient requires accurate assessment of the prolapse lead point in each case. Often, this can be done on physical exam alone. Persistent or recurrent cases of prolapse should prompt further investigative studies. Potter recommends evaluation of the rectum with a barium contrast study or flexible sigmoidoscopy prior to surgery to evaluate the lead point and rule out solitary rectal ulcers [31]. Findings of megarectum or megacolon on plain films will require resection of the dilated bowel; neither sclerotherapy nor rectopexy will be sufficient in these cases.

With a high success rate and low complication and recurrence rates, we recommend injection sclerotherapy as the initial intervention for pediatric rectal prolapse. Due to the simplicity and efficacy of the laparoscopic rectopexy, we advocate this procedure to be the next attempted for persistent or recurrent rectal prolapse. Intestinal

resection should be reserved for patients with rectal or sigmoid redundancy. More morbid procedures should rarely be required.

3.2. Study limitations

Our review has several important limitations. Most of the studies included in this review are case series reported from single institutions. Publication and selective reporting biases are therefore highly likely. Authors may have hand-picked cases to publish, and complication rates may be under-reported. Additionally, many of the case series had only a small number of patients included. Patient demographics and prior interventions differed across each case series, and some patients may have had rectal prolapse secondary to another condition. Each case series utilized different protocols, standards of evidence, and follow-up schedules. Because we cannot control for these sources of bias, we have attempted to mitigate these effects by not performing a rigorous statistical analysis of our results. Instead, we sought to present a comprehensive review that includes the best available evidence and to evaluate that evidence critically and holistically instead of statistically. Additionally, we have looked outside the realm of pediatric rectal prolapse literature to bring in supplemental data that can strengthen our understanding of potential complications. We hope this evaluation will guide the reader in their clinical decision-making. Nonetheless, results reported in this review should be interpreted with caution.

4. Conclusion

Treatment of pediatric rectal prolapse is initially nonoperative with manual reduction and bowel regimens. We recommend that procedural intervention for persistent prolapse begin with injection sclerotherapy, and we have found ethyl alcohol to be a highly efficacious and readily-available sclerosing agent. On failure of injection sclerotherapy, the laparoscopic rectopexy is a minimally-invasive transabdominal procedure with a low complication rate that can often be performed on an outpatient basis. With high reported success rates, more morbid procedures should rarely be necessary.

Declarations of interest

None.

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