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Efficacy of Enema via Cecostomy for Fecal Disorders in Children: A Systematic Review and Meta-Analysis

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Abstract

Background
Some controversy exists about the role of cecostomy in the management of fecal disorders. The present meta-analysis aims provide a comprehensive evaluation on the role of cecostomy on management of fecal incontinence and constipation in children.

Materials and Methods
An extensive search was performed on the Medline, Embase, Scopus, and Web of Science until July 2017. Two independent researchers screened the title and abstracts of the studies and then relevant studies were included. Finally, pooled effect size was presented as standardized mean difference (SMD) or pooled prevalence with 95% confidence interval (95% CI).

Results
14 articles were entered (740 children). The success rate of cecostomy in management of fecal disorders was 90.6% (success rate=90.6%; 95% CI: 86.4 to 94.2). The most common side effects of this technique include granulation tissue formation (29.6%), cecostomy tube leakage (8.5%), tube dislodgement (7.0%), and tube site infection (2.3%).

Conclusion
The results of the present meta-analysis show that the cecostomy is safe and an effective technique in the management of fecal disorder in children. However, the findings presented on the eligible articles have have shown a low level of evidence and it is suggested that clinical trials should be conducted in this field.

Key Words: Antegrade Colonic Enema, Constipation, Fecal Incontinence, Pediatrics.

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1- INTRODUCTION

The normal structure and sensorimotor function of the internal and external sphincter are essential for fecal continence (1). The fecal incontinence and constipation are the prevalent pelvic floor disorders. The prevalence of these disorders is reported to be between 0.4% and 22.0% (2-5). These disorders cause social stigma, mental problems, and ultimately depression and isolation of the patient that greatly affects the quality of life (6). The incidence of complications in children is higher than in adults (7). The general belief is that surgery is the most effective therapeutic strategy in treatment of fecal incontinence. However, recently a systematic review has shown that there is no sufficient evidence to support the use of surgical treatments (8). Therefore, it is necessary to use less invasive methods such as laxative, bulking agents, manual evacuation, enema and biofeedback (9).

In addition to the conventional methods, recent studies suggest the novel strategies such as cell therapy and neuromodulation (10-13). One of these non-invasive methods, which has received considerable attention in recent years, is continence enemas (CE) through cecum known as cecostomy (14). In this method, the appendix has been used as a catheterization stoma for CE. Cecostomy leads to patient's independence, increases self-esteem and improves quality of life (7). There is some controversy about the role of cecostomy in the management of fecal incontinence and constipation.

One of the ways to achieve a conclusion is to perform a systematic review. Since the 1990s, when Malone et al proposed cecostomy for management of fecal disorders (15), many efforts have been made to assess safety and efficacy of cecostomy. Accordingly, the present systematic review and meta-analysis aims provide a comprehensive conclusion on the role of cecostomy on management of fecal incontinence and constipation in children.

2- MATERIALS AND METHODS

2-1. Search strategy

The present study was conducted based on the Cochran's guidelines. An extensive search was performed on the Medline, Embase, Scopus, and Web of Science until July 2017. The search query in Medline (via PubMed) is shown in **Table.1**. In addition, a manual search was conducted in Google motor engine, Google Scholar, and bibliography of related articles and reviews.

Table-1: Search strategy for Medline (via PubMed).

Search terms
("Fecal Incontinence"[Mesh] OR "Fecal Incontinence"[tiab] OR "Fecal Leak"[tiab] OR "Faecal Incontinence"[tiab] OR "Fecal Incontinences"[tiab] OR "Defecation Disorder*"[tiab]) AND ("Cecostomy"[MeSH] OR "Cecostomy"[tiab] OR "Cecostomies"[tiab] OR "Tube Cecostomy"[tiab] OR "Cecostomies, Tube"[tiab] OR "Tube Cecostomies"[tiab] OR "Antegrade colonic enemas"[tiab] OR "Antegrade Continence/Colonic Enema"[tiab] OR " Antegrade continence enemas"[tiab]).

2-2. Selection criteria

Studies that evaluated the effects of cecostomy on management of childhood fecal incontinence and constipation were included. No time and language limitations were applied. Studies that have used combination therapy and review articles were excluded.

2-3. Quality Assessment and Data Extraction

The method of data collection, summarizing of data and quality control of eligible studies have been reported in previous studies (16-22). Briefly, after conducting the search and eliminating duplicated reports, two independent researchers screened the title and abstracts of the studies and then potentially relevant studies were selected. Any disagreement was solved by discussion. Data related to the design of the study, characteristics of patients (age, sex), sample size, type of disorder (fecal incontinence and constipation), follow-up duration, outcomes, and probable bias were recorded. Outcomes were categorized into two general categories. The first was the success rate and the second was post-intervention complications including granulation tissue formation, infection, leakage, tube dislodgment and other complications. The quality of the studies was assessed using Cochrane's proposed guidelines (23).

2-4. Statistical analysis

Statistical analyses were performed by STATA version 14.0 (Stata Corporation, College Station, TX). Heterogeneity was assessed using I^2 statistics, and p value less than 0.1 was considered as significant (representing heterogeneity). Fixed effect model (if the studies were homogeneous) and random effect model (if the heterogeneity was observed) was used and pooled effect size was presented as standardized mean difference (SMD) or pooled prevalence with 95% confidence interval (95% CI). In addition, Egger's and Begg's tests were used to assess the publication bias (24).

3- RESULTS

3-1. Summary of included studies

Systematic and manual search led to achieving 1328 non repetitive records. After the screening, 14 articles were entered (1, 25-37) (**Figure.1**). Except for one study that had case- control design (26) the rest of the studies were case-series design. 740 children were included. Among them 531 cases had fecal incontinence, 179 cases had constipation, and 30 patients have both. Cecostomy protocol was explained completely in all studies. Follow up duration was between 8 days and 18 years (**Table.2**) (*Please see the Table.2 in the end of paper*).

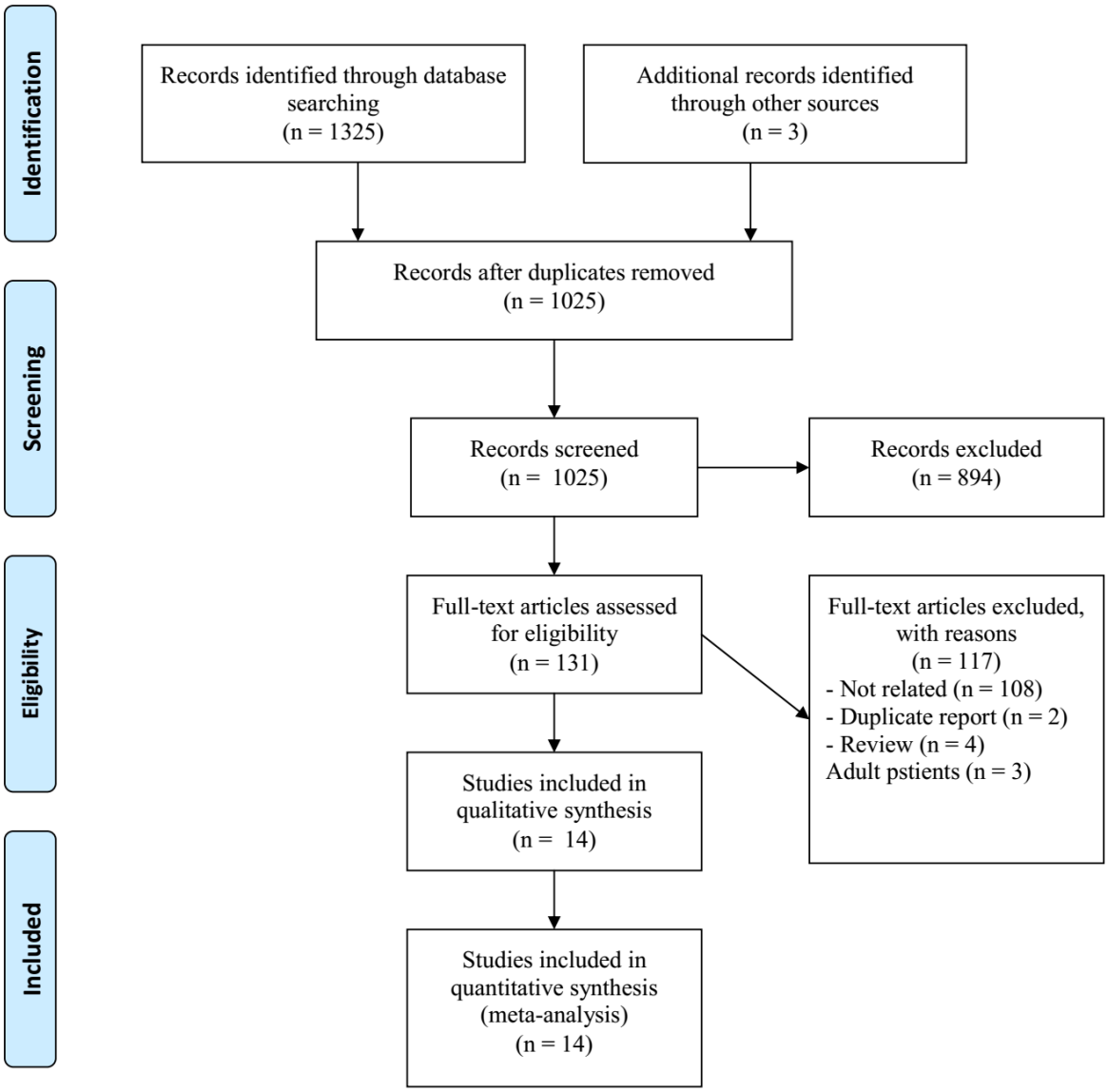

Fig1: PRISMA flowchart of present meta-analysis.

3-2. Risk of bias

The risk of bias in blinding of patients and researchers, blinding of outcomes assessment, random sequence generation and other bias were high. Risk of bias of selective reporting in all included studies was unclear (**Figure.2**). Publication bias was not observed among eligible articles (bias coefficient= 0.99, p=0.39).

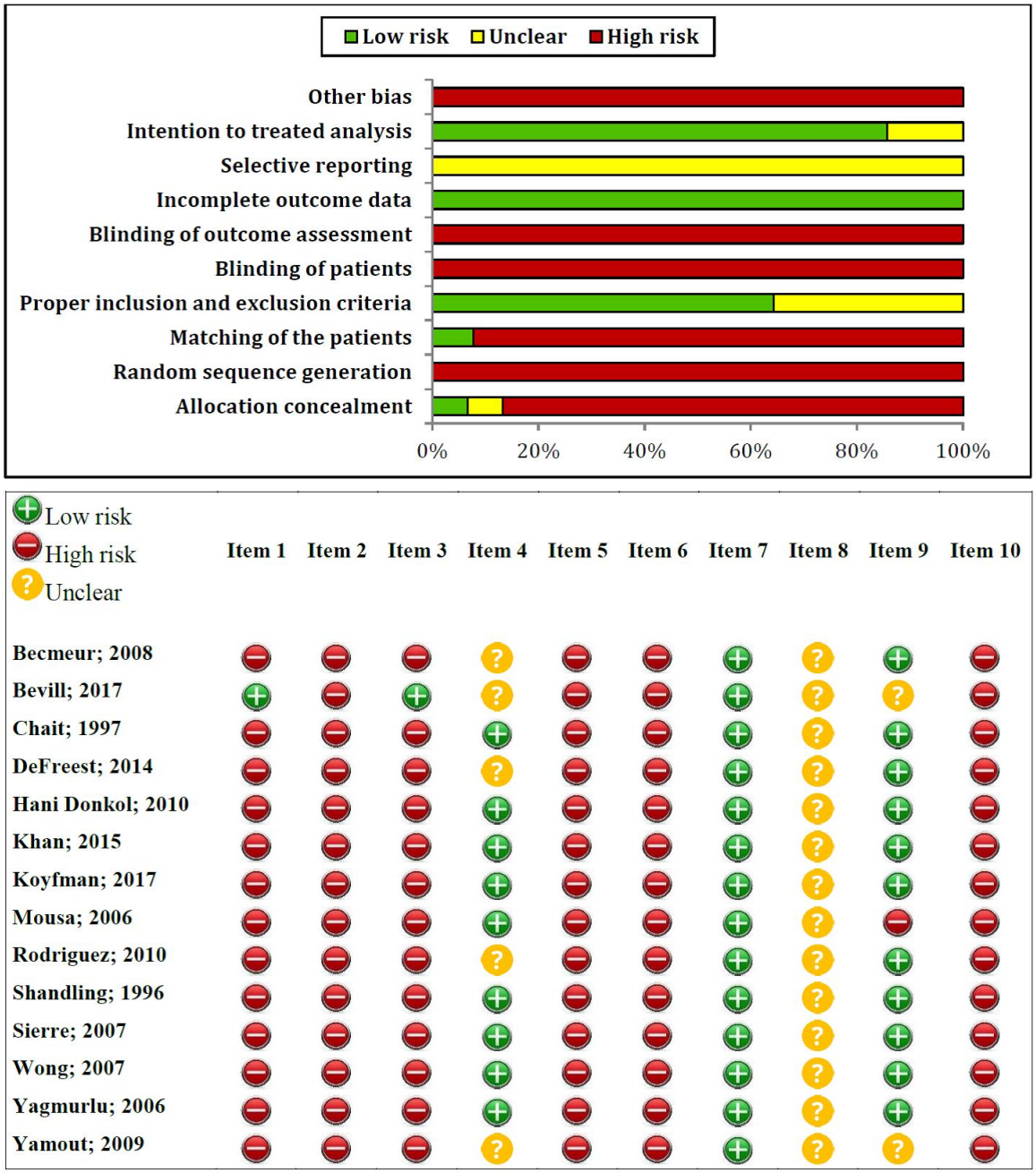

Fig.2: Quality control of eligible studies. Item 1: Allocation concealment; Item 2: Random sequence generation; Item 3: Matching of the patients; Item 4: Proper inclusion and exclusion criteria; Item 5: Blinding of patients; Item 6: Blinding of outcome assessment; Item 7: Incomplete outcome data; Item 8: Selective reporting; Item 9: Intention of treated analysis; Item 10: other bias.

3-3. Meta-analysis

3-3-1. Success rate

10 studies were attempted to investigate the success rate for cecostomy in fecal incontinence (1, 25-28, 30, 31, 33, 34, 36). Pooled analysis showed that the success rate of cecostomy in management of fecal disorders was 90.6% (success rate=90.6%; 95% CI: 86.4 to 94.2). In this part heterogeneity did not exist (I^2=0.0%, p>0.99) (**Figure.3**).

3-3-2. Complications

3-3-2-1. Granulation tissue formation

Data of 11 article (447 children) were entered in the analysis (1, 25-28, 30, 31, 33, 35, 37). The results showed that heterogeneity did not exist in this part (I^2=0.0%, p>0.99). As **Figure.4** depicts the prevalence of granulation tissue formationafter cecostomy is 29.6% (SMD= 29.6%, 95% CI: 24.6 to 34.8).

3-3-2-2. Tube site infection

Data of 11 articles comprising 656 children were entered in the analysis (1, 26-32, 34, 35, 37). The pooled analysis showed that the prevalence of infection after cecostomy is 2.3% (SMD= 2.3%, 95% CI: 1.1 to 3.9; I^2=0.0%, p>0.99) (**Figure.4**).

3-3-2-3. Cecostomy tube leakage

Data of 9 articles with the sample of 332 children were entered in the analysis (25-27, 30-32, 34, 35, 37). Pooled analys.s showed that 8.5% (SMD= 8.5%, 95% CI: 5.5 to 12.0) of patients had leakage around cecostomy tube site (I^2=0.0%, p>0.99) (**Figure.4**).

3-3-2-4. Tube dislodgement

Tube dislodgement was another issue that was evaluated in 11 articles (1, 27-32, 34-37). This part of analysis includes 610 children. The prevalence of tube dislodgement after cecostomy treatment was 7.0% (SMD= 7.0, 95% CI: 4.9 to 9.5; 12=0.0%, p>0.99) (**Figure.4**).

3-3-2-5. Other complication

Other complications were reported in 6 articles (26, 29, 32, 34, 35, 37) which include 514 children (I^2=0.0%, p>0.99). As **Figure.4** shows the prevalence of this issue is 7.3% (SMD= 7.3, 95%CI: 5.1 to 9.9) (**Figure.4**).

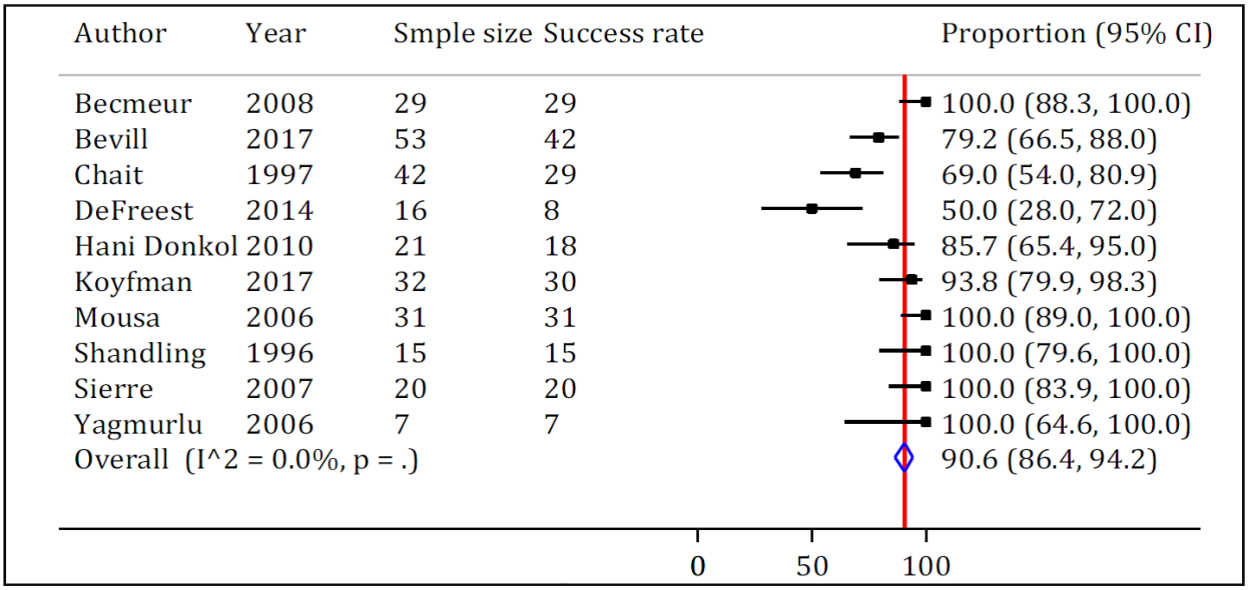

Fig.3: Success rate (continence important) of cecostomy in management of fecal disorders. CI: Confidence interval.

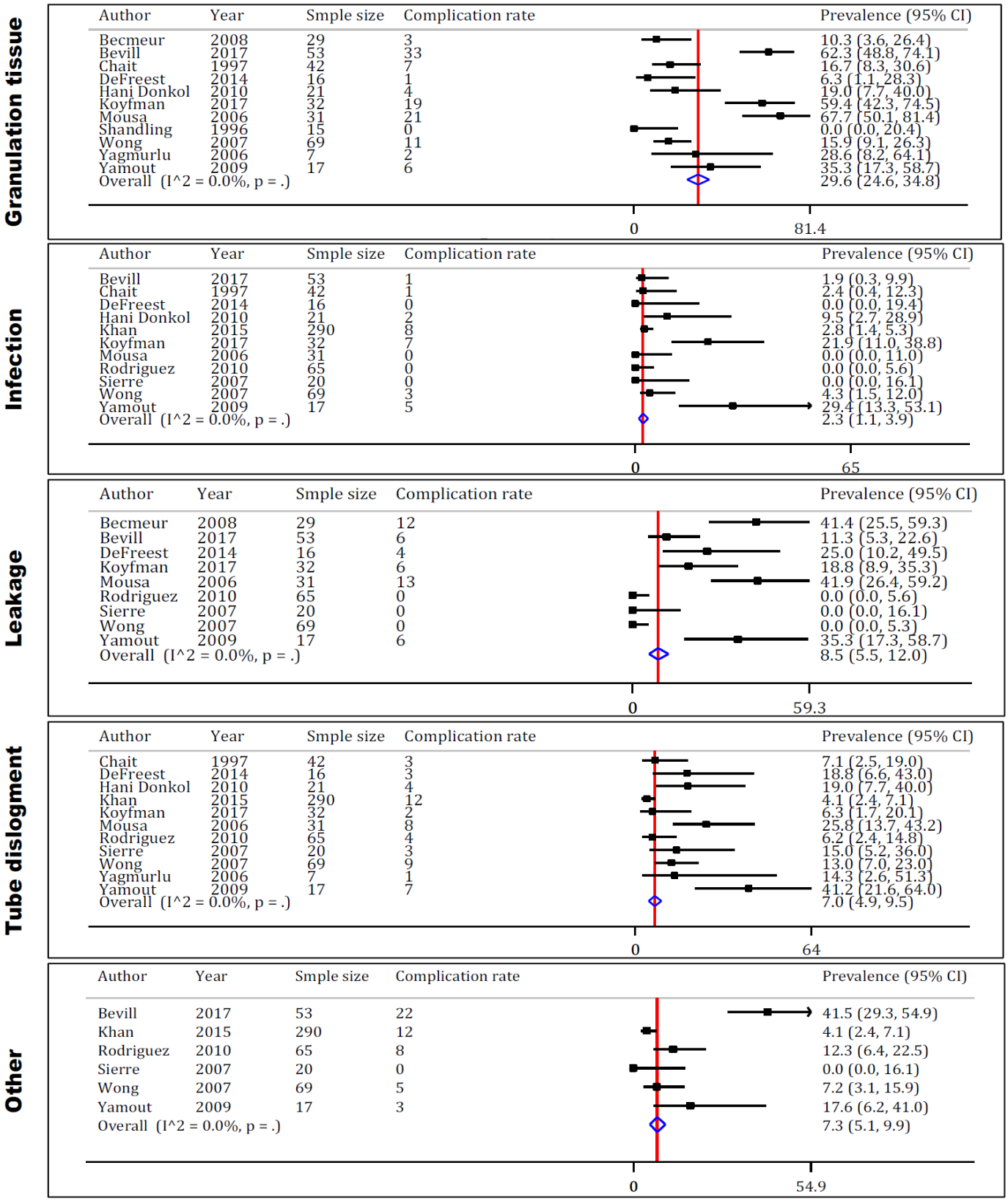

Fig.4: Complications rate of cecostomy in management of fecal disorders. Ci: Confidence interval.

4- DISCUSSION

For many years, the cecostomy has been done for various purposes. For the first time, in 1986, Casola et al., introduced cecal catheterization through the skin to remove pressure in the adult colon. They stated that this method is efficient in fecal disorders (38). In 1998, Ganc et al., showed that transcolonoscopic extraperitoneal cecostomy is an effective therapeutic technique for management of fecal incontinence (39). The results of the present meta-analysis indicate the high success rate of the cecostomy in treatment of pediatric constipation and fecal incontinence. The most common side effects of this technique include granulation tissue formation, tube site leakage, tube dislodgement, and infection.

In children with fecal disorders, non-surgical treatments include changing dietary habits, the use of laxatives, bulking agents, and retrograde colonic enemas (15, 40). The success rate of retrograde colonic enemas is low due to colon and perineal muscle weakness (41). So, the idea of antegrade colonic enema was first introduced in 1990 by Malone and colleagues. They showed that antegrade colonic enema (MACE) can be used to control constipation and fecal incontinence (15). Since the patients can determine the colon emptying time with the use of MACE, they remain completely clean (27). During MACE, antegrade flows through the cecostomy leads to complete colon emptying due to access to the entire colon. This mechanism is different from retrograde enema because in retrograde method access to all sections of colon is not possible, especially the transverse and ascending colon. As a result, complete emptying of the colon is not possible and the probability of fecal disorder recurrence at intervals between sequential enemas is high (42). In addition, during MACE the person is in the sitting position on the toilet and feels privacy and comfort (27). In 2016, Chan et al., conducted a systematic review and meta-analysis to examine the role of antegrade continence enema in controlling fecal disorders in adults. In this study, 17 articles (426 adult patients) were included. They reported that success rate was 74.3% (95% CI: 66. 1 to 82.6) and the most common complication was wound infection (22 ± 8%) and stomal stenosis (16.6%) (43). In our study, the success rate for MACE in the children was 90.6%, and the most common complications were granulation tissue formation (29.6%), tube leakage (8.5%), and tube dislodgment (7.0%), respectively. Based on the findings of this research, the success rate of cecostomy in children is high in the control of fecal disorders.

However, the success rate of invasive procedures, such as surgery depending on the type of disorder and the definition of success rate, is between 38% and 80% (44-50). In addition, side effects of cecostomy are most commonly less complicated than surgical technique (51). The complication rates observed in this study range from 2.3 to 29.6%. However, in the surgery, the observed complication rates vary between 20% and 87% (52-61).

4-1. Limitation

92.8% of eligible studies have no control group and their design has been case-series. Therefore, conducting well-designed clinical trials is necessary. In addition, in most studies, the role of cecostomy in the quality of life has not been assessed.

5- CONCLUSIONS

The results of the present systematic review and meta-analysis show that the cecostomy is safe and effective in management of fecal disorder in children. However, the findings presented on the eligible articles have shown a low level had a low level of evidence. Therefore, it

is suggested that clinical trials should be conducted in this field.

6- CONFLICT OF INTEREST: None.

7- ACKNOWLEDGMENTS

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Table-2: Summary of included studies.

Author; Year; Country	Type of study	Sample size	Number of patients*	Age range (year)	No. male	Population	Follow up duration	Outcome
Becmeur; 2008; France	Case-series	29	24 / 5 / 0	3 to 21	18	Fecal incontinence or encopresis and constipation	NR	Success rate and complications
Bevill; 2017; USA	Case-Control	86	0 / 86 / 0	3 to 42	38	Neurogenic bowels	NR	Success rate and complications
Chait; 1997; Canada	Case-series	42	42 / 0 / 0	2 to 20	23	Fecal incontinence	8 to 503 days	Success rate and complications
DeFreest; 2014; USA	Case-series	16	1 / 15 / 0	6 to 16	8	Fecal incontinence and constipation	6 to 51 months	Success rate and complications
Hani Donkol; 2010; France	Case-series	21	21 / 0 / 0	5 to 16	13	Fecal incontinence	12 to 23 months	Success rate and complications
Khan; 2015; Canada	Case-series	290	290 / 0 / 0	3 to 18	170	Fecal incontinence	3 to 18 years	Complications
Koyfman; 2017; USA	Case-series	32	1 / 31 / 0	0 to 19	18	Fecal incontinence or encopresis and constipation	0 to 71.9 months	Success rate and complications
Mousa; 2006; USA	Case-series	31	22 / 9 / 0	3 to 18	18	Fecal incontinence and constipation	11 months	Success rate and complications
Rodriguez; 2010; USA	Case-series	65	4 / 31 / 30	NR	43	Fecal incontinence or encopresis and constipation	32 to 37 months	Complications
Shandling; 1996; Canada	Case-series	15	15 / 0 / 0	7 to 20	NR	Fecal incontinence	NR	Success rate and complications
Sierre; 2007; Argentina	Case-series	20	20 / 0 / 0	6 to 12	8	Fecal incontinence	27.6 months	Success rate and complications
Wong; 2007; canada	Case-series	69	69 / 0 / 0	NR	NR	Fecal incontinence	120 months	Complications
Yagmurlu; 2006; USA	Case-series	7	5 / 2 / 0	4 to 12	3	Fecal incontinence and constipation	15 months	Success rate and complications
Yamout; 2009; USA	Case-series	17	17 / 0 / 0	5 to 17	8	Fecal incontinence	4 to 67 months	Complications

*, data are presented as incontinence / constipation / both; NR: Not reported.