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Review Article

A Systematic Review and Meta-Analysis of Comparing Drainage Alone versus Drainage with Primary Fistula Treatment for the Perianal Abscess in Children

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Abstract

This systematic review and meta-analysis of nonrandomized studies (NRSs) aimed to evaluate the clinical efficacy and safety of two types of surgical interventions (respectively drainage alone and drainage with primary fistula treatment) for perianal abscesses (PAs) in children. Studies from 1992 to July 2022 were searched in 10 electronic databases. All relevant NRSs with available data which compared surgical drainage with or without primary fistula treatment were included. Patients with underlying diseases which led to abscess formation were excluded. The Newcastle-Ottawa Scale was used to assess the risk of bias and quality of the included studies. The outcomes were the healing rate, fistula formation rate, fecal incontinence, and wound healing duration. A total of 16 articles with 1,262 patients were considered suitable for the final meta-analysis. Primary fistula treatment was associated with a significantly higher healing rate when compared with incision and drainage alone (odds ratio [OR]: 5.76, 95% confidence interval [CI]: 4.04–8.22). This aggressive procedure for PA resulted in an 86% reduction in the fistula formation rate (OR: 0.14, 95% CI: 0.06–0.32). Limited data showed patients who underwent primary fistula treatment have a minor effect on postoperative fecal incontinence. Primary fistula treatment demonstrates a better clinical efficacy in promoting the healing rate and decreasing the formation of fistulas in PAs in children. The available evidence for a minor impact on anal function after this intervention is less strong.

Keywords

- systematic review
- meta-analysis
- perianal abscess
- children
- primary fistula treatment

Introduction

A perianal abscess (PA) is an acute inflammatory condition mainly characterized by a collection of pus.[1] It has been previously reported that only a small proportion of patients are children.[2–4] In recent years, an increasing amount of evidence suggests that these diseases are no longer considered rare conditions in childhood. Patients are often accompanied by acute and severe pain originating from erythema, swelling, induration, and fluctuance in the anal area, whereas disseminated systemic infection or sepsis are unusual.[5]

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Fistula-in-ano (FIA) is an epithelialized connection between perianal skin and the anorectal canal, which represents chronic manifestation developed from the PA. It is widely accepted that abscesses arise from the obstruction of anal glands.[6] Additionally, several predisposing disorders may also be responsible for the development of an abscess, such as inflammatory bowel disease (IBD), anorectal malformations, and immunodeficiency.[7,8]

With a clear predominance of males, most children have the onset of PA in the first year of life.[9,10] Various therapies have been applied, while decision-making on choosing the treatment depends on the experiences and judgments provided by physicians, as well as the tendency of caregivers. Nonoperative management will be more acceptable due to its safety and efficacy, while prompt incision and drainage (I&D) still should be implemented for abscesses with significant fluctuation, which provoke pain or systemic signs of sepsis and cannot discharge spontaneously.[11,12] Yet, the rate of fistula formation after this initial management among studies was reported to be around 10 to 30%.[10,13–15]

Some surgeons suggest that I&D with primary fistula treatment (I&DF) can reduce the possibility of recurrence and progression to FIA. However, such a procedure may also be associated with a risk of fecal incontinence or transient manometric reduction in anal sphincter pressures in previous meta-analyses on adults.[16,17] For children, it is also affiliated with concerns about the safety of anesthesia exposure.[18] This controversial topic has not been deeply explored so far in this special population.

Our preliminary searches revealed that there were few randomized controlled trials (RCTs) focusing on this issue, and all of the relevant studies were from China which might cause a potential source of bias and addressed the review question incompletely in children.[19–26] According to the Cochrane Handbook for Systematic Reviews of Intervention, we decided to consider nonrandomized studies (NRSs) for objectively evaluating the benefits and harms of these two surgical interventions among the pediatric population.[27] Thereby, an extensive search of the literature was conducted to reduce the possibility of publication bias. The aim of this systematic review and meta-analysis which compare outcomes after I&DF and I&D was to determine the role of primary fistula treatment in efficacy and complications.

Methods

This systematic review and meta-analysis were registered with the number CRD42022331529 (https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42022331529).

Criteria Used to Consider Studies for This Review

Type of studies: Nonrandomized clinical trials from 1992 through July 2022 which compared outcomes after I&DF and I&D for PAs in children were included. Published articles in any available language were considered. Nonclinical trials, clinical trials with no control, or RCTs were excluded.

Type of participants: Children (< 18 years old) undergoing surgical drainage of PAs without the diagnosis of IBD, anorectal malformations, Hirschsprung's disease, tuberculosis, malignancy, and immunosuppressive disorders.

Type of interventions: Two types of interventions studied were compared with each other:

Treatment group (I&DF): I&DF, which was comprised of fistulotomy, fistulectomy, and use of cutting setons, as well as addressing the offending crypt and identifying an internal opening.

Control group (I&D): I&D alone.

Search Methods for Study Identification

The following international databases were searched: PubMed, Embase, Cochrane Central Register of Controlled Trials (CENTRAL), Scopus, and Web of Science. In addition, the Chinese databases were also searched, including China National Knowledge Infrastructure (CNKI), VIP, Wanfang Data, Sinomed, and Yiigle.

To achieve comprehensive search results relevant to the study topic, there was no restriction on the study type. Three of the authors (Y.S., S.H., and X.Z.) conducted the systematic literature search, using a combination of the following terms: "anal," "abscess," "child," and "drainage." Correlative subject headings, free words, synonyms, along with wildcards were used to expand the retrieval scope. The search strategy used to search PubMed is shown in ►**Supplementary Table S1**. The search strategies were modified to adapt different electronic databases.

All duplications were precluded through identification in Endnote. Articles, as follows, were also kept out through title/abstract screening by two authors (Y.S. and X.Z.) independently: (1) irrelevant to the study topic, (2) patients with other underlying diseases, which had been listed above, (3) a review or meta-analysis, (4) a case report or meeting abstract, and (5) an in vitro study or animal experiment. The secondary screening was based on a full-text assessment to determine whether studies met the inclusion criteria. Studies without sufficient information were also excluded from the meta-analysis. In case of disagreement, a third reviewer (S.H.) was involved to reach a consensus. The selection process was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 statement.[28]

Data Extraction and Selection

Three authors independently (Y.S., S.H., and X.Z.) extracted data from each included study into prepared datasheets. When encountering inconsistencies, we arbitrated through discussion with the corresponding author (C.W.). The first primary outcome was the healing rate defined as the complete healing of the fistula after the operation by clinical assessment. Another primary outcome was the fistula formation rate during the follow-up period. The secondary outcomes were fecal incontinence and wound healing duration, which represented the number of days taken for complete closure of the wound. Other variables about characteristics and treatment details of included NRSs were also collected. Any data missing or unavailable in the published articles were marked as not reported (NR).

Risk of Bias Assessment

The Newcastle-Ottawa Scale was used to assess the quality of included cohort NRSs by two authors separately (S.H. and X.Z.). The final results were determined by discussing with two other authors (Y.S. and C.W.). Each study, which can be awarded from 0 to 9 stars, was evaluated from three broad perspectives using the above-mentioned method. A higher score indicated a high quality of the selected article. The study quality can be classified into three grades: low (scores < 5), moderate ($5 \leq$ scores < 8), and high ($8 \leq$ scores ≤ 9). The final score of a study, which was equal to or greater than 5, was selected for further meta-analysis.

Strategy for Data Synthesis

Relevant variables were extracted and saved in a Microsoft Excel datasheet. Results for dichotomous data are expressed as numbers (proportion), whereas continuous variables are presented as mean and standard deviation (SD). The meta-analysis was conducted using Review Manager 5.4.1 software. Pooled odds ratio (OR) with 95% confidence intervals (CI) was estimated for dichotomous outcomes (healing rate and fistula formation rate), and mean difference with 95% CI were calculated for continuous outcomes (wound healing duration). Cochrane Q test and I^2 statistics were used to assess the heterogeneity among studies, which was accepted when p-value greater than 0.1 and I^2 less than 50%. If p-value less than 0.1 or I^2 greater than 50%, which suggested a higher probability of inconsistency, the random-effects model was applied; otherwise, the fixed-effect model was conducted. Publication bias was assessed through the funnel plot when the number of included studies was more than 10.

Results

Literature Search

Overall, 3,933 records were retrieved from 10 electronic databases between 1992 and July 2022. After the exclusion of duplicates, 79 articles were left through primary screening of the title and abstract. Ultimately, 16 NRSs were eligible for the final meta-analysis. Eight studies were retrospective,[12,29–35] 7 were prospective,[36–42] and 1 was NR.[43] Among these, 11 articles were in Chinese, and 5 studies in English were from New Zealand, Sweden, Australia, the United Kingdom, and China, respectively. A flow diagram of the search and selection process is shown in ►Fig. 1.

Study Characteristics

We presented the characteristics of 16 included NRSs in ►Table 1. There were a total of 1,262 patients, including 1,066 males and 196 females. All participants included in the

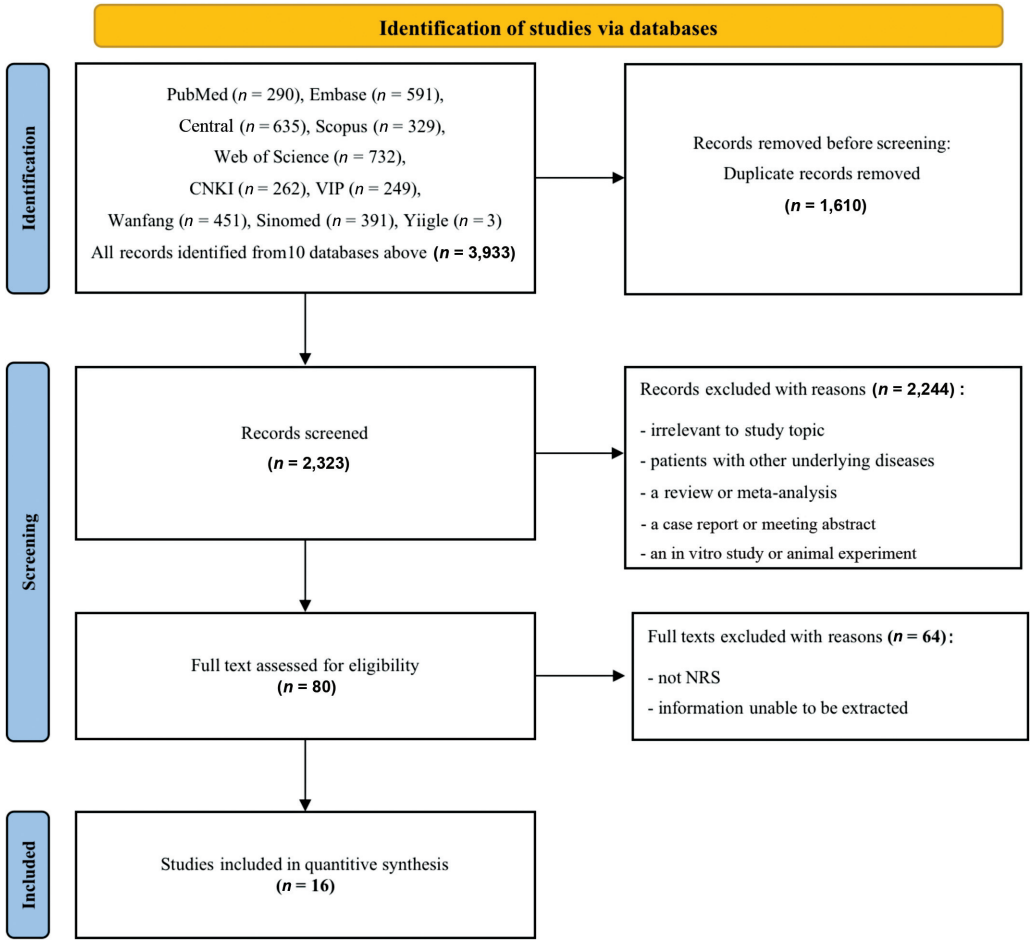

Fig. 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) diagram showing search results for meta-analysis.

Table 1 Characteristics of included NRSs

Study	Year	Country	Total number of patient	Male	Treatment group intervention	Group		Age Mean (min, max)	Disease duration Mean ± SD or (min, max)	Follow-up Mean (min, max)
						Treatment	Control			
Tian et al[43]	2001	China	30	19 (63.33%)	Drainage with fistulotomy	15	15	25 d, 8 y	5 d, 1 mo	NR
Murthi et al[29]	2002	UK	33	29 (87.88%)	Drainage with fistulotomy	20	13	Male: 10.5 mo (1 mo, 17 y) Female: 7.5 y (6 y, 13 y)	NR	3 y (1 y, 5 y)
Xu[36]	2007	China	102	89 (87.25%)	Drainage with cutting seton	52	50	T: 18.3 d (5 d, 28 d) C: 21.1 d (7 d, 27 d)	T: 5.5 ± 2.5 d C: 4.5 ± 2.5 d	3 y
Kuang[30]	2007	China	31	24 (77.42%)	Drainage with fistulotomy	6	25	NR	3 d, 6 d	4 mo, 18 mo
Buddicom et al[31]	2012	New Zealand	91	85 (93.41%)	Drainage with fistulotomy	66	25	25 m (1 mo, 16 y)	NR	NR (patients from October 1996 to April 2009)
Xiao[37]	2012	China	53	51 (96.23%)	Drainage with fistulotomy	25	28	T: 28 d (8 d, 32 d) C: 26 d (7 d, 28 d)	2 d, 6 d	6 mo, 2 y
Wang et al[38]	2015	China	112	69 (61.61%)	Drainage with fistulotomy	56	56	T: 3.8 y (30 d, 5 y) C: 3.9 y (31 d, 5 y)	T: 25.9 ± 2.5 d, (1 d, 90 d) C: 26.4 ± 3.5 d (1 d, 91 d)	2 mo
Juth Karlsson et al[32]	2016	Sweden	84	80 (95.24%)	Drainage with fistulotomy	34	50	5 m (8 d, 15 y)	NR	3 y (6 mo, 7 y)
Yu and Lei[33]	2017	China	30	30 (100%)	Drainage with cutting seton	7	23	NR	NR	> 2 mo
Zeng[39]	2017	China	146	127 (86.99%)	Drainage with fistulotomy	98	48	T: 17 d, 5 y C: 18 d, 5 y	T: 2 d, 17 d C: 2 d, 18 d	NR
Chen[40]	2017	China	76	42 (55.26%)	Drainage with fistulotomy or cutting seton	38	38	T: 5.85 y (7 mo, 11 y) C: 6.3 y (6 mo, 12 y)	T: 2.4 d ± 0.3 d C: 2.2 d ± 0.5 d	NR
Yao[34]	2018	China	102	97 (95.10%)	Drainage with fistulotomy	53	49	< 28 d	NR	6 mo
You and Zhang[41]	2019	China	30	15 (50%)	Drainage with fistulotomy or cutting seton	15	15	T: 31.43 d C: 30.23 d	NR	NR
Chen et al[42]	2019	China	96	65 (67.71%)	Drainage with fistulotomy or cutting seton	50	46	T: 3.67 y (30 d, 6 y) C: 3.49 y (30 d, 6 y)	T: 4.37 ± 1.63 d C: 4.38 ± 1.57 d	3 mo

Table 1 (*Continued*)

Study	Year	Country	Total number of patient	Male	Treatment group intervention	Group		Age Mean (min, max)	Disease duration Mean ± SD or (min, max)	Follow-up Mean (min, max)
						Treatment	Control			
Tan Tanny et al[12]	2020	Australia	108	107 (99.07%)	Drainage with fistulotomy	85	26	T: 66.5 d (21 d, 328 d) C: 41.5 d (18 d, 332 d)	NR	NR (patients over a 10-year period)
Yin et al[35]	2022	China	138	137 (99.28%)	Drainage with fistulotomy	65	73	T: 22.71 d, < 28 d C: 22.36 d, < 28 d	T: 4 d (2 d, 5.5 d) C: 3 d (2 d, 5 d)	6.5 y (4 y, 13 y)

Abbreviations: NR, not reported; NRSs, nonrandomized studies; SD, standard deviation.
Note: T represents the treatment group and C represents the control group.

control group had undergone I&D of PAs, while those in the treatment group received I&DF. In one study, 108 patients with 111 abscesses were divided into two groups.[12] Finally, 685 cases were enrolled in the treatment group, and 580 cases were drained alone. In detail, the majority of patients were managed by drainage with fistulotomy while cutting setons were selectively employed for PAs with the high location of identified internal openings.[40–42] Patients in two studies were all performed by drainage with cutting setons.[33,36] The age of patients, disease duration, and follow-up period was described in a variety of ways. The youngest age of patients in the included studies was all less than 28 days.[34,35] All reported disease durations were less than 30 days in 8 studies,[30,35–37,39,40,42,43] and just 1 article demonstrated the longest duration was 91 days.[38] The follow-up in most studies was longer than 6 months, and the longest one was 13 years.[35]

Further treatment information of included NRSs was demonstrated in ►**Table 2**. The anesthetic approaches comprised general and non-general anesthesia, including caudal and local anesthesia with or without conscious sedation.[30,32,33,35,38–40] Intravenous anesthesia was performed as one of the anesthetic techniques of administration.[39] Ketamine was used for inducing anesthesia, which was considered a way of basal anesthesia.[36] The locations of abscesses were mostly lateral to the anus according to the existing information, and a more detailed description was presented at 3 and 9 o'clock in the lithotomy position.[12,32,33,35–37] Five articles clearly stated the step of searching for fistulas before allocation. A lacrimal probe was introduced to identify the fistula in four articles,[12,29,31,32] and the remaining one article reported the methods of digital anal examination and anoscopy.[40] The internal openings were either laid open or dealt with cutting setons. Laying open with diathermy down onto the probe was provided in a detailed description of the surgical procedure.[12,31,32] Setons, used in the fashion of cutting, were placed through the fistula tract.[33,36,40–42] Antibiotics, which were in oral, topical, or intravenous use, varied in the relevant studies.[33,35,40] Some patients received preoperative or postoperative antibiotics, and some were treated in conjunction with the surgeries.[12,29,32,36–38,41]

Risk of Bias Assessment

Overall, the results of the methodological quality assessment of the 16 studies were satisfactory, which are summarized in ►**Supplementary Table S2**. Four of them were assessed as high-quality,[35–37,42] and all the remaining were considered of moderate quality.[12,29–34,38–41,43] There was only one study that had no description of the derivation of the nonexposed cohort.[43] All studies controlled for age based on design or analysis, which was deemed the most important factor. Each study could be awarded 2 stars when other factors were also considered, such as the disease duration.[33,35–40,42,43] Outcomes in eight studies were identified via record linkage.[12,29–33,35,42] The follow-up period (equal to or more than 6 months) was regarded as long enough for outcomes to occur.[29,32,34–37]

Table 2 Summary of treatment details of included NRSs

Study	Mode of anesthesia	Abscess location	Searching for fistulas before allocation	Surgical procedure for treating internal opening in detail	Antibiotic use
Tian et al[43]	NR	0.3–1 cm from the anal margin	No	Lay open	NR
Murthi et al[29]	NR	NR	Yes (except 6 patients)	Lay open	Some patients received preoperative antibiotics
Xu[36]	Ketamine anesthesia	Lateral to the anus, 0.5–1.5 cm from the anal margin	No	Cutting seton	Postoperative antibiotics use
Kuang[30]	General anesthesia	~1 or 2 cm from the anal margin	No	Lay open	NR
Buddicom et al[31]	NR	NR	Yes (except 2 patients)	Lay open with diathermy dissection down onto the probe	NR
Xiao[37]	NR	Lateral to the anus	No	Lay open	Postoperative antibiotics use
Wang et al[38]	T: basal anesthesia C: local, basal anesthesia or without anesthesia	NR	No	Lay open	Postoperative antibiotics use
Juth Karlsson et al[32]	General anesthesia	Lateral to the anus in most cases	Yes (except 35 patients)	Treating with monopolar diathermy to the probe	Some patients received antibiotic therapy in conjunction with surgical treatment
Yu and Lei[33]	Local anesthesia	Lateral to the anus	No	Cutting seton	Topical antibiotics
Zeng[39]	Local or intravenous anesthesia	NR	No	Lay open	NR
Chen[40]	T: caudal anesthesia C: anesthesia	NR	Yes	Lay open or cutting seton	T: topical antibiotics C: None
Yao[34]	NR	NR	No	Lay open	NR
You and Zhang[41]	Anesthesia	NR	No	Lay open or cutting seton	Postoperative antibiotics use
Chen et al[42]	Basal anesthesia	NR	No	Lay open or cutting seton	NR
Tan Tanny et al[12]	NR	Lateral to the anus	Yes	Lay open with diathermy down onto the probe	Preoperative antibiotics use
Yin et al[35]	T: conscious sedation and local anesthesia C: local anesthesia	Lateral to the anus in most cases mainly at 3 and 9 o'clock	No	Lay open with diathermy	Intravenous antibiotics use

Abbreviations: NR, not reported; NRSs, nonrandomized studies.
Note: T represents the treatment group and C represents the control group.

Outcome Parameter

An overview of the primary outcomes and secondary outcomes is tabulated in ►**Table 3**. Outcomes with sufficient data available were eligible for further meta-analysis.

Healing rate: The healing rate in the treatment group ranged from 60 to 100%, and most were above 90% except for three studies.[12,41,42] In the control group, almost all the studies demonstrated a healing rate of less than 90%, and the least was 40%.[41] Only 1 trial described that 21 of 23 participants were cured by I&D alone, revealing a higher healing rate of 91.30%.[33] Since no conspicuous heterogeneity appeared among the studies, the fixed-effect model was employed for merging and calculating the pooled OR and 95% CI. The pooled results indicated that there was a

Table 3 Outcome variables in the analyzed NRSs

Study	Number of PAs	Healing rate		Fistula formation		Fecal incontinence		Wound healing time (d) Mean ± SD or estimation	
		Treatment	Control	Treatment	Control	Treatment	Control	Treatment	Control
Tian et al[43]	30	14/15 (93.33%)	10/15 (66.67%)	1/15 (6.67%)	5/15 (33.33%)	NR		NR	
Murthi et al[29]	33	20/20 (100%)	6/13 (46.15%)	0/20 (0%)	5/13 (38.46%)	NR		NR	
Xu[36]	102	49/52 (94.23%)	21/50 (42.00%)	NR		NR		24.5 ± 3.5	28.5 ± 4.5
Kuang[30]	31	6/6 (100%)	22/25 (88.00%)	0/6 (0%)	3/25 (12%)	NR		NR	
Buddicom et al[31]	91	61/66 (92.42%)	19/25 (76.00%)	NR		NR		NR	
Xiao[37]	53	25/25 (100%)	15/28 (53.57%)	0/25 (0%)	6/28 (21.43%)	NR		NR	
Wang et al[38]	112	55/56 (98.21%)	49/56 (87.50%)	NR		NR		12.6 ± 1.9	20.5 ± 0.9
Juth Karlsson et al[32]	84	31/34 (91.18%)	30/50 (60.00%)	NR		NR		NR	
Yu and Lei[33]	30	7/7 (100%)	21/23 (91.30%)	NR		NR		NR	
Zeng[39]	146	96/98 (97.96%)	39/48 (81.25%)	NR		NR		12.7 ± 1.8	20.6 ± 1.1
Chen[40]	76	35/38 (92.11%)	27/38 (71.05%)	0/38 (0%)	3/38 (7.89%)	1	2	NR	
Yao[34]	102	51/53 (96.23%)	41/49 (83.67%)	NR		NR		16.8 ± 2.9	15.1 ± 2.6
You and Zhang[41]	30	9/15 (60.00%)	6/15 (40.00%)	1/15 (6.67%)	5/15 (33.33%)	NR		NR	
Chen et al[42]	96	31/50 (62.00%)	19/46 (41.30%)	2/50 (4%)	5/46 (10.87%)	Wexner score: T < C anal manometry: T > C		NR	
Tan Tanny et al[12]	111	72/85 (84.71%)	17/26 (65.38%)	NR		NR		NR	
Yin et al[35]	138	64/65 (98.46%)	59/73 (80.82%)	1/65 (1.54%)	10/73 (13.70%)	0	0	21	14

Abbreviations: NR, not reported; NRSs, nonrandomized studies; PAs, perianal abscesses; SD, standard deviation.
Note: T represents the treatment group and C represents the control group. Anorectal manometry: Resting pressure and maximum squeezing pressure.

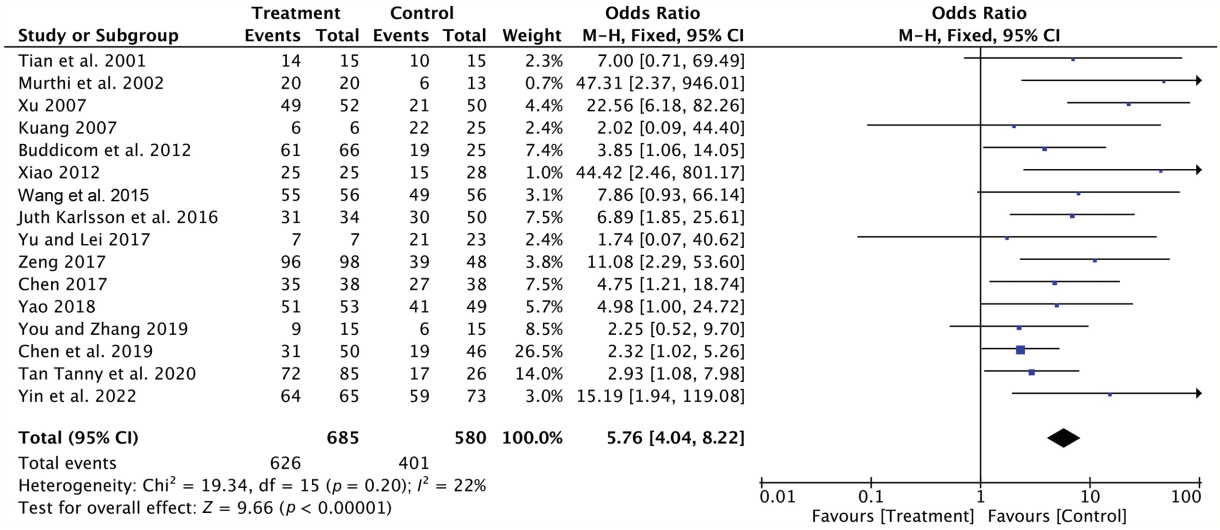

Fig. 2 Forest plot of healing rate for the treatment group versus the control group. CI, confidence interval; I&DF, treatment, incision, and drainage with primary fistula treatment; I&D, control, incision, and drainage.

significant difference between the treatment group and the control group (OR: 5.76, 95% CI: 4.04–8.22, $I^2 = 22\%$, $p < 0.001$) (►**Fig. 2**). The funnel plot did not suggest any obvious publication bias (►**Supplementary Fig. S1**).

Fistula formation rate: Eight out of 16 studies had reported cases of fistula formations during the time of study or follow-up period.[29,30,35,37,40–43] None of the patients in the treatment group developed a fistula in 4 trials,[29,30,37,40] and the highest formation rate was 6.67% in the 2 articles.[41,43] The fistula formation rates were remarkably higher in the control group, which varied from 7.89 to 38.46%. There was no evidence of heterogeneity among these relevant studies. Primary fistula treatment of PAs in children gave rise to an obvious reduction in the risk of fistula formation after surgical drainage (OR: 0.14, 95% CI: 0.06–0.32, $I^2 = 0\%$, $p < 0.001$) (►**Fig. 3**).

Fecal incontinence: Fecal incontinence was only reported in three Chinese studies, two of which documented the number of patients, and the other presented a comparison of preoperative and postoperative anal manometry combined with the Wexner Incontinence Score.[35,40,42] There was a transient decrease in anal resting pressure and maximum squeeze pressure, along with a slight increase in the Wexner score 1 month after surgery in both groups. This situation had changed and recovered after 3 months postoperatively, and the treatment group had significantly more improvement than the control group had in the Wexner score.[42] No other information was available on the consequence of fecal incontinence in the remaining articles.

Wound healing time: Five articles with 600 patients showed the wound healing time, among which 4 were reported as mean ± SD,[34,36,38,39] and 1 recorded estimated time.[35] Three of them indicated that the average time for wound healing was shorter in the treatment group than that in the control group,[36,38,39] and the opposite situation was observed in two articles.[34,35] There was also a discrepancy in the length of time between both groups. For instance, the shortest mean duration was 12.6 days and the longest was 24.5 days in the treatment group. Due to the large degree of heterogeneity, it was not possible to carry out a quantitative meta-analysis to compare the efficacy of two types of interventions in wound healing time.

Discussion

PAs and fistulas represent different entities of the same disease process, indicating the acute and chronic

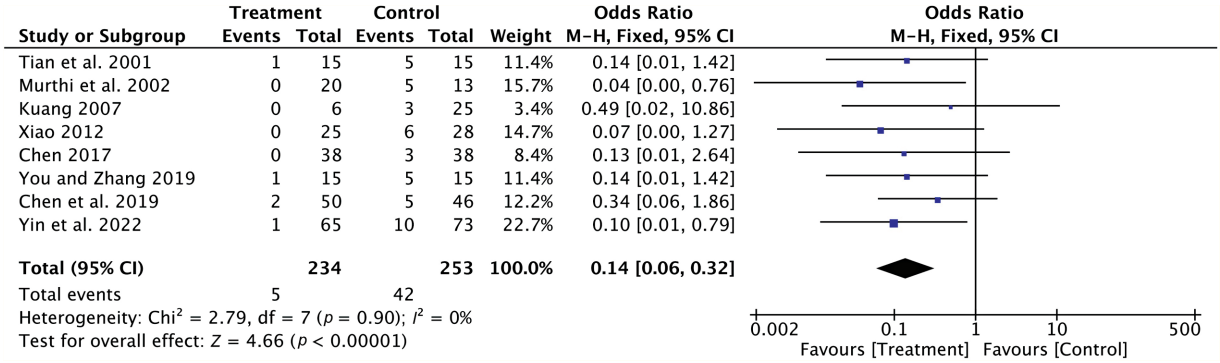

Fig. 3 Forest plot of fistula formation rate for the treatment group versus the control group. CI, confidence interval; I&DF, treatment, incision, and drainage with primary fistula treatment; I&D, control, incision, and drainage.

inflammatory phases of perianal infection. Although the disease primarily affects adults, it has also been known to afflict children for many years. I&D is the mainstream surgical treatment with the advantage of a fast, simple, and efficient procedure. For most abscesses with superficial fistulas, primary fistula treatment has been previously advocated to prevent the recurrence of PAs and the development of fistulas based on the theory of cryptoglandular origin.[44,45] This aggressive surgical approach may increase the likelihood of fecal incontinence, so it remains controversial.

To the best of our knowledge, this is the first systematic review and meta-analysis for assessing postoperative outcomes and relevant information between I&DF and I&D in children with PAs. Our major objective was to evaluate the clinical efficacy and safety of both surgical approaches. The pooled results revealed a significant elevation in the healing rate for patients with I&DF. Of 1,265 included abscesses, 626 (91.39%) in the treatment group versus 401 (69.14%) in the control group illustrated complete solutions with no further intervention over the follow-up period according to all 16 NRSs. The odds of fistula formation rate in children treated with I&DF was 86% lower than those treated with I&D. Among eight included articles with available data, four studies reported no incidence of progression to fistulas in the treatment group.[29,30,37,40] Regarding the results of our meta-analysis, the positive effects of primary fistula treatment seemed to be more pronounced than those of drainage alone. Concerning the wound healing time, patients treated with I&DF appeared to recover more quickly in three studies.[36,38,39] But the differences were too large to carry out the pooled analysis, which contributed to the considerable heterogeneity. We speculated that several factors might influence the inconsistency, including the size of abscesses and surgical incisions, age variation, and postoperative care.

Yet, there was very little evidence of fecal incontinence and no consensus had been reached on the assessment in children. Only three studies from China, among which one was written in English, mentioned the postoperative anal function of patients.[35,40,42] Although the surgery might have an impact on the anal sphincter, the results revealed less possibility of incontinence and better preservation of anal function in the treatment group than that in the control group. In addition, another three English articles indicated that a vast majority of fistulas with relatively superficial positions involved less or no sphincter in children and thus the risk of incontinence was supposed to be small.[12,31,32] This might be the reason why investigators did not conduct the follow-up on fecal incontinence. From our perspective, children tend to have a high level of limb activity, making it challenging to conduct quiet and cooperative examinations such as anal manometry and perianal magnetic resonance imaging. Moreover, different patterns of bowel movements can impact the assessment of the severity of fecal incontinence. This also poses a difficulty in establishing a standardized evaluation process for anal sphincter function in the pediatric population. Thus, longer follow-ups are warranted to determine whether the effects persist into adulthood or not.

It has been demonstrated that coexisting fistulas are present in approximately 60 to 76% of children at the initial treatment of PAs.[12,29,31,32] Examinations for fistulas were not applied in all cases due to the potential harm to exploration and the risk of false opening and iatrogenic tract.[46] Nevertheless, primary fistula treatment was indeed considered to be beneficial for children to decrease the likelihood of recurrences and avoid secondary surgery.[47] The key to improving efficacy and safety depends on identifying the accurate location of internal openings contiguous with fistulas, as well as figuring out the amount of anal sphincter involved. More detailed preoperative examinations are necessary, such as transcutaneous perianal ultrasonography.[48] That also requires skilled and experienced surgeons with sufficient knowledge of anatomy, who will probe the corresponding anal crypt gently but thoroughly to look for a fistula. On the other hand, the abnormal deep and thick crypts of Morgagni have been recognized as a cause of cryptitis in children, which can easily develop into a PA.[49] Therefore, I&DF will enable to prevent the persistent infections at the origin of the defects.[50] Usually, the associated fistulas are simple intersphincteric or subcutaneous types and locate superficially with less penetration through muscles in children, especially in infants.[14,51–53] For internal openings in high positions, cutting setons are more favored to minimize the damage to the anal function. There have been few reports about the functional or anatomical impact of this surgical procedure on the anal sphincter in children currently.[54] So, we are unable to demonstrate that primary fistula treatment should be advocated under all circumstances.

All the included studies with nonrandomized designs and limited data might have potential bias and confounding factors, which could affect the results. Among these studies, 12 trials were from China and 4 were from other countries. The variation of the study population and a broad spectrum of ages could be the reason for the disparity in treatment outcomes. Not all the studies had documented whether these abscesses were first-time presentations or recurrent abscesses. The available disease durations were almost less than 2 weeks, while only 1 article reported an average time of approximately 25 days.[38] This suggested the possibility of bias since a longer duration or recurrence was more likely to be accompanied by an underlying fistula. Consequently, those patients might be more inclined to receive primary fistula treatment. The processes of searching for fistulas during surgery were explicitly mentioned only in 5 studies, among which 43 patients did not receive the search for fistulas.[12,29,31,32,40] Patients who had undergone this important procedure were also more likely to be allocated to the treatment group. Antibiotics, which had been implemented in different ways, were applied in 10 trials.[12,29,32,33,35–38,40,41] Some believed antibiotic administration showed benefits for PAs in children while others found no effect on the recurrences.[12,29,32] It was hard to say what could be the role of antibiotics in treating abscesses, but this was really a possible confounding factor that might influence the results.

Our systematic review and meta-analysis especially focused on pediatric patients who suffered from PAs and were

treated by surgical interventions (I&DF and I&D). The results suggested that primary fistula treatment was considered beneficial at the time of I&D, which might improve the healing rate and decrease the likelihood of fistula formation. Although there was insufficient evidence to support that such an aggressive procedure did not cause apparent injuries to anal sphincters, we speculated the superficial location of abscesses and protective treatments for high internal openings were associated with minor postoperative fecal incontinence. A thorough and gentle search for fistulas by skilled and experienced surgeons intraoperatively is demanded to avoid excessive tissue damage and preserve anal function. Well-established assessment of postoperative anal function and detailed research information will be necessary to provide strong and supportive evidence. We would like to revisit this issue when more useful data from multicenter, randomized, double-blind clinical trials of high quality can be obtained in the future.

Data Availability

All data generated or analyzed during this study are included in this published article and its supplementary information files.

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Conflict of Interest

None declared.

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