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ORIGINAL ARTICLE

One-stage repair of anorectal malformations in females with vestibular fistula: a systematic review and meta-analysis

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Abstract

Purpose Females with recto-vestibular fistula (RVF) can be managed either by one-stage sagittal anorectoplasty (SARP) or by conventional multi-stage approach with colostomy followed by SARP. Our aim was to define which approach, one-stage or multi-stage, is safer and more beneficial.

Methods Using a defined search strategy, two investigators identified all comparative studies on the mentioned procedures. The study was conducted under PRISMA guidelines. The meta-analysis was performed using RevMan 5.3. Data are mean ± SD.

Results Of 649 titles/abstracts screened, 13 full-text articles were analyzed. Three studies were included (156 females). One-stage SARP was associated with increased risk of wound infection (24.3 ± 8.7%) compared to multi-stage approach (10.9 ± 2.5%; $p < 0.01$) and increased risk of wound dehiscence (16.2 ± 4.8% vs. 2.4 ± 1.1%, respectively; $p < 0.01$). The incidence of anorectal stenosis was higher following one-stage repair (33.3%) vs. multi-stage approach (10.7%; $p < 0.05$). No differences were found with regards to redo SARP in both groups (12.9 ± 7.3% vs. 4.8 ± 0.8%; p = ns). At follow-up, the prevalence of soiling and constipation were similar after one-stage (19.7 ± 10.3% and 29.5 ± 5.4%) and multi-stage repair (13.7 ± 8.9% and 28.7 ± 4.4%; p = ns).

Conclusions In females with RVF, the SARP performed without protective colostomy increases the risk of postoperative complications. However, this one-stage approach seems not to be associated with reduced fecal continence.

Keywords Anorectal malformations · Recto-vestibular fistula · One-stage sagittal anorectoplasty · Systematic review · Meta-analysis

Introduction

Anorectal malformations occur in 1 in 5000 births and affect boys and girls equally. Recto-vestibular fistula (RVF) is the most common type of anorectal malformations in newborn girls [1]. Posterior sagittal anorectoplasty (PSARP) described by Peña and anterior sagittal anorectoplasty (ASARP) introduced by Okada are the most commonly used surgical techniques [2, 3]. However, females with RVF could be managed either by a single-stage definitive repair or conventional two or three-staged approach [4]. One-stage sagittal anorectoplasty (SARP) is a definitive repair without prior colostomy creation. However, this exposes the child to increased risk of wound dehiscence and therefore to a greater risk of damage to the sphincteric mechanism owing to acquired severe fibrosis [5]. Therefore, a covering colostomy is still considered the safest way to avoid these complications. The staged repair is either a three-step approach (colostomy, SARP, and colostomy closure) or a two-stage repair characterized by (1) colostomy formation and subsequent SARP with concomitant closure of the colostomy or (2) SARP plus colostomy as the first procedure and colostomy closure as the second step. Although a

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colostomy avoids infection complications and dehiscence [6], it is associated with many complications, including prolapse, retraction, skin excoriation, stricture, and adhesive obstruction, which are poorly tolerated by both young children and parents [7, 8]. The question of whether these disadvantages outweigh the protective effect of a colostomy from wound infection and dehiscence following SARP is still controversial.

The aim of the present study was to define which approach, one-stage or multi-stage repair is safer and more beneficial in the surgical repair of RVF in females.

Materials and methods

Both the systematic review and the meta-analysis were drafted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [9]. Two different health librarians were involved at the Gerstein Science Information Centre (University of Toronto, ON, Canada) and the Bibl@Ud'A (University of Chieti-Pescara, Italy).

Systematic review

The present study was registered on PROSPERO—international prospective register of systematic reviews (registration number: CRD42018109160) [10]. A systematic review of the literature was made using a defined search strategy. Two investigators (GL and DDR) independently searched scientific databases (PubMed, Medline, Cochrane Collaboration, Embase and Web of Science) using a combination of keywords (Table 1). MeSH headings and terms used are "Recto-vestibular fistula", "Recto-vestibular fistula AND sagittal ano-rectoplasty", and "Recto-vestibular fistula AND one-stage sagittal ano-rectoplasty" (Supplementary file 1). Case reports, opinion articles, experimental studies and case series with less than ten patients were excluded. All grey literature publications (i.e., reports, theses, conference proceedings, bibliographies, commercial documentations, and official documents not published commercially) were excluded. The full text of the potentially eligible studies was retrieved and independently assessed for eligibility by the same two investigators. Any disagreement between them over the eligibility of particular studies was resolved through discussion with a third author (AZ).

Meta-analysis

Only studies comparing one-stage SARP vs. multi-stage SARP (two or three procedures) were included. Outcome measures included the incidence of wound infection, wound dehiscence, rescue colostomy after one-stage approach, anorectal stenosis, redo SARP in both groups. At long-term follow-up, outcome measures included the prevalence of soiling and constipation. The meta-analysis was conducted with RevMan 5.3 [11], using the random-effects model to produce risk ratio (RR) for categorical variables and mean differences (MD) for continuous variables, along with 95% confidence intervals (CI). We produced I^2 values to assess homogeneity and quantify the dispersion of effect sizes.

Data were compared using Fisher's exact test and are expressed as mean ± SD. When median and range were reported, mean ± SD were estimated, as previously reported [12].

Table 1 Inclusion criteria of systematic review

Publication	
Language	Any
Date	After 1950
Subject	Human studies
Study type	Retrospective
	Prospective
	Case control
	Cohort
Excluded	Case reports
	Case series
	Letters
	Editorials
	Grey literature
Keywords	Anorectal malformations
	Recto-vestibular fistula
	Posterior sagittal anorectoplasty
	Anterior sagittal anorectoplasty
	One-stage repair

Quality assessment

Risk of bias for individual studies was assessed in duplicate (GL and DDR) using the methodological index for non-randomized studies (MINORS) [13]. Differences between the two reviewers (GL and DDR) were resolved through consensus and discussion with another author (AZ). The total score for this 12-item instrument ranges 0–24 points with a validated "gold standard" cut-off of 19.8. Two authors (PLC and AP) independently evaluated the present systematic reviews and meta-analysis using A Measurement Toll to Assess Systematic Reviews (AMSTAR) [14]. The PRISMA checklist of our study was then completed [9].

Results

Systematic review

Of 649 titles/abstracts screened, 13 full-text articles were analyzed. Six studies on one-stage SARP met our inclusion criteria (199 females, Fig. 1) [15–20]. Of these papers, five were retrospective [15–17, 19, 20] and one was a randomized controlled trial (RCT) [18]. Moreover, three papers reported results with a posterior approach (PSARP) [15, 18, 19] and three studies included patients treated with a supine position (ASARP, Table 2) [16, 17, 20].

Minor wound infections were reported in 27/199 cases (13.6%) and wound dehiscence was present in 13/173 patients (7.5%). When mentioned, the incidence of anorectal

Table 2 Studies included in the systematic review

Author	Year	Type of study	Type of SARP	Type of staged SARP
Menon et al. [15]	2007	R	PSARP	n.r
Upadhyaya et al. [16]	2007	R	ASARP	n.r
Wang et al. [17]	2015	R	ASARP	n.r
Amanollahi et al. [18]	2016	RCT	PSARP	3-stage
Karakus et al. [19]	2017	R	PSARP	3-stage
Khalifa et al. [20]	2018	R	ASARP	2-stage

R retrospective, *RCT* randomized controlled trial, *SARP* sagittal anorectoplasty, *PSARP* posterior sagittal anorectoplasty, *ASARP* anterior sagittal anorectoplasty, *n.r.* not reported

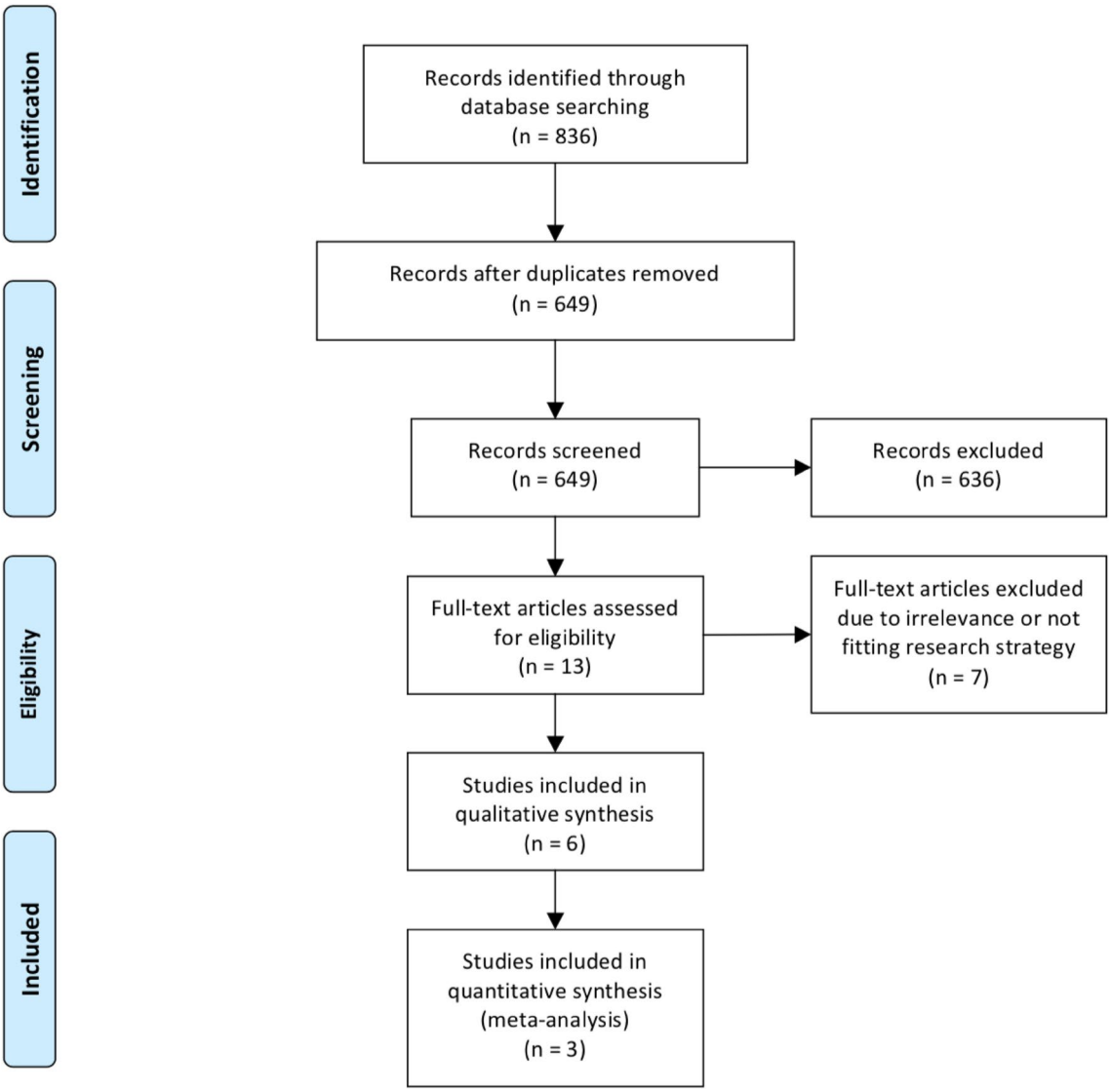

Fig. 1 Diagram of workflow in the systematic review and meta-analysis

stenosis was 6.6% (8/122 cases), anal retraction has been reported in 3/96 patients (3.1%), and rectal prolapse in 6/148 females (4.0%). The recurrence of RVF with subsequent redo SARP has been reported in 3/177 cases (1.7%).

At long-term follow-up, constipation was reported in 27/186 females (14.5%) and soiling was present in 15/186 cases (8.1%).

Meta-analysis

Three studies comparing one-stage SARP vs. staged SARP were included (156 females, 1 RCT, Table 3) [18–20]. Two of these papers compared one-stage PSARP vs. three-stage approach [18, 19] and one study compared one-stage ASARP vs. two-stage approach (with a simultaneous sigmoid colostomy and ASARP at the first stage) [20] (Table 2). One-stage SARP was associated with an increased risk of wound infection (18/74, 24.3 ± 8.7%) compared to multi-stage approach (9/82, 10.9 ± 2.5%; $p < 0.01$, RR 2.76, 95% CI 1.33–5.73, $I^2 = 0\%$; Fig. 2a) and with an increased risk of wound dehiscence/disruption (12/74, 16.2 ± 4.8% vs. 2/82, 2.4 ± 1.1%, respectively; $p < 0.01$, RR 5.80, 95% CI 1.73–19.48, $I^2 = 0\%$; Fig. 2b). Due to this complication, a rescue colostomy was necessary in 8/74 patients in the one-stage group (10.8 ± 7.3%). Conversely, complications of colostomy such as stenosis, prolapse, and infection have been reported in 12/66 cases in the staged group (18.2 ± 1.1%).

The incidence of anorectal stenosis was higher following one-stage repair (8/54, 14.8 ± 8.3%) vs. multi-stage approach (5/62, 8.1 ± 2.7%; $p < 0.05$, RR 3.07, 95% CI 1.13–8.36; Fig. 3a). No differences were reported in both groups with regards to rectal prolapse (5/50, 10.0 ± 2.1% vs. 5/36, 13.9 ± 6.2%, respectively; p = ns, RR 0.76, 95% CI 0.23–2.49, $I^2 = 0\%$; Fig. 3b) and anal retraction (3/24, 12.5% vs. 1/46, 2.2%, respectively; p = ns, RR 5.75, 95% CI 0.63–52.35; Fig. 3c). Moreover, no differences were found between one-stage repair and multi-stage approach with regards to redo SARP (7/54, 12.9 ± 7.3% vs. 3/62, 4.8 ± 0.8%, respectively; p = ns, RR 1.45, 95% CI 0.04–49.10, $I^2 = 76\%$; Fig. 3d).

At long-term follow-up, the prevalence of voluntary bowel movement did not differ in one-stage group (32/41, 78.0 ± 6.8%) in comparison with staged SARP (45/60, 75 ± 5.8%; p = ns, RR 1.00, 95% CI 0.85–1.18, $I^2 = 0\%$; Fig. 4a). The prevalence of soiling was similar after one-stage (12/61, 19.7 ± 10.3%) and multi-stage repair (11/80, 13.7 ± 8.9%; p = ns, RR 1.32, 95% CI 0.66–2.64, $I^2 = 0\%$; Fig. 4b). Finally, constipation did not differ between these two groups (18/61, 29.5 ± 5.4% vs. 23/80, 28.7 ± 4.4%, respectively; p = ns, RR 1.12, 95% CI 0.67–1.86, $I^2 = 0\%$; Fig. 4c).

Discussion

This systematic review and meta-analysis indicates that children with RVF who had one-stage repair compared to multi-stage repair have an increased incidence of wound complications (wound infection, dehiscence, and anorectal stenosis). However, this increased incidence of complications does not affect their future fecal continence and constipation [19].

Table 3 Studies included in the meta-analysis

	Amanollahi et al. [18]		Karakus et al. [19]		Khalifa et al. [20]	
	1-stage 20 pts (%)	3-stage 20 pts (%)	1-stage 30 pts (%)	3-stage 16 pts (%)	1-stage 24 pts (%)	2-stage 46 pts (%)
Wound infection	4 (20)	1 (5)	2 (6.7)	1 (6.2)	10 (41.7)	7 (15.2)
Wound dehiscence/disruption	2 (10)	0 (0)	3 (10)	0 (0)	7 (29.2)	2 (4.3)
Rescue colostomy	1 (5)	–	n.r	–	7 (29.2)	–
Colostomy complications	–	3(15)	–	n.r	–	9 (19.5)
Anorectal stenosis	n.r	n.r	0 (0)	0 (0)	8 (33.3)	5 (10.9)
Rectal prolapse	3 (15)	5 (25)	2 (6.7)	0 (0)	n.r	n.r
Anal retraction	n.r	n.r	n.r	n.r	3 (12.5)	1 (2.1)
Redo SARP	n.r	n.r	0 (0)	1 (6.2)	7 (29.2)	2 (4.3)
Length follow-up	Range 11–35 months		Till pts toilet trained		Till pts toilet trained	
Voluntary bowel movement	n.r	n.r	16/17 (94.1)	13/14 (92.9)	16 (66.7)	32 (69.6)
Soiling	0 (0)	0 (0)	7/17 (41.2)	5/14 (35.7)	5 (20.8)	6 (13.0)
Constipation	4 (20)	3 (15)	4/17 (23.5)	5/14 (35.7)	10 (41.7)	15 (32.6)

SARP sagittal anorectoplasty, *n.r.* not reported

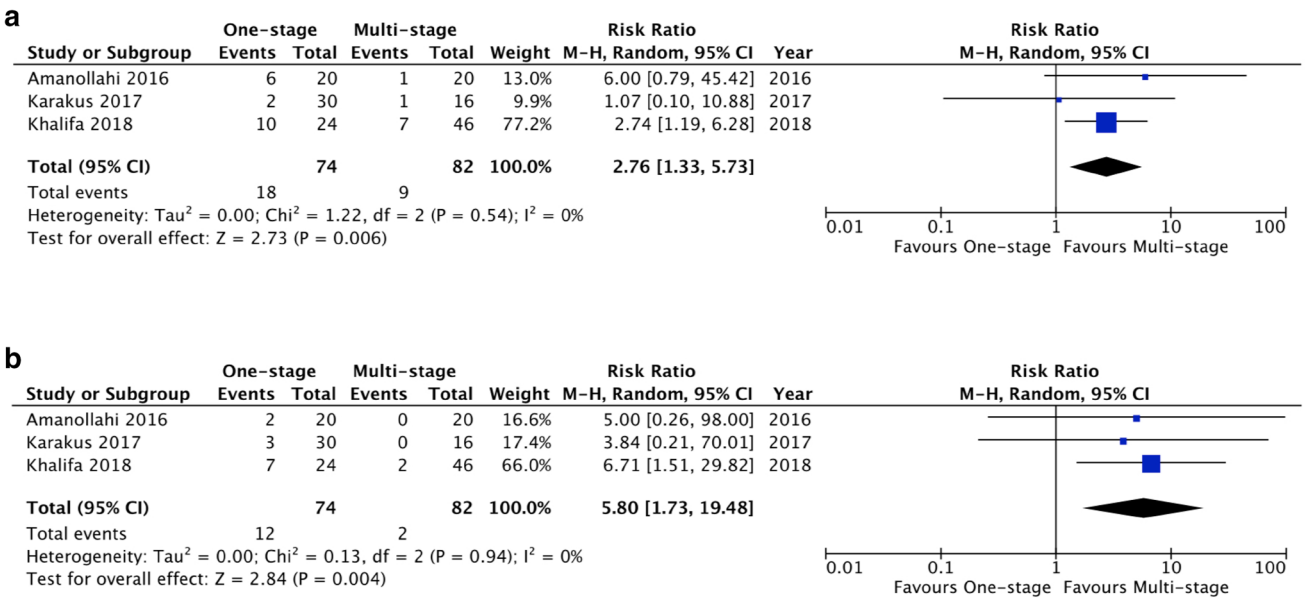

Fig. 2 **a** Comparison of the incidence of wound infection between one-stage repair versus multi-stage approach; **b** comparison of the incidence of wound dehiscence/disruption

Many procedures has been described to manage females with RVF, including cutback operation, V–Y plasty, anal transposition, PSARP, and ASARP [8, 9]. The PSARP provides excellent exposure of the RVF and a precise placement of the rectum within the striated muscle complex giving best chance to achieve continence [21]. However, the PSARP involves division of external sphincter, muscle complex, and part of the lower portion of the levator mechanism [10]. Various authors claimed that ASARP has many advantages over PSARP, such as an easier mobilization of rectum from vagina under direct vision, the avoidance of levator ani division, and an accurate reconstruction of both sphincteric muscle and perineal body [20, 22]. ASARP involves division of the anterior fibers of the external sphincter.

Selecting the multi-stage or one-step surgical procedure to treat this malformation has been a subject of debate for many years [18]. Levitt and Peña published a flow chart, which describes the indications for a colostomy or primary repair [23]. However, the ARM-Net consortium indicates that the size of RVF is a clinical factor to be considered in the decision of performing a repair with or without a protective colostomy [24]. These authors suggested a primary repair or colostomy in females with small RVF (Hegar size < 5 mm), and delayed or primary repair without colostomy in patients with larger RVF (Hegar size > 5 mm) [24].

The main reason for choosing the multi-stage repair is the small risk of wound infection and wound dehiscence following fecal diversion by colostomy [7, 8, 21, 25, 26]. These complications would compromise the expected good functional outcome in this type of anomaly [15, 27]. Moreover, the colostomy allows performing the repair a few months later when the child is slightly heavier, making the operation potentially easier. On the other hand, the reasons for choosing one-stage repair are multiple: avoidance of multi-stage operations, saving time and costs, and less stress and insult for children and their parents. Furthermore, anorectoplasty without colostomy means avoidance of colostomy-related complications, reported up to 70% in the literature ($18.2 \pm 1.1\%$ in our systematic review) [3, 15, 25, 28–31]. However, Peña et al. attribute the high colostomy complication rate to technical inadequacies in stoma formation and stoma management. They emphasized that the high stoma-related complications should not be considered as a supportive argument for doing a primary definitive procedure without protective colostomy [7].

Wound infection and wound dehiscence are potential complications of SARP. The virtually sterile meconium during the first week of life should reduce the risk of infection from fecal contamination [18]. In addition, it has been suggested that wound contamination could be minimized by aggressive cleansing of the bowel by pre-operative total bowel irrigation and by keeping the patient nil per oral for the first five postoperative days [5, 15, 19]. Similarly, the use of perioperative therapeutic antibiotics seems to be associated with decreased wound infection. Few studies have reported a lower incidence of wound infection after postoperative antibiotics (up to 7 days) compared to children who had no antibiotics or prophylactic antibiotics (24 h) [16, 17, 25, 32, 33].

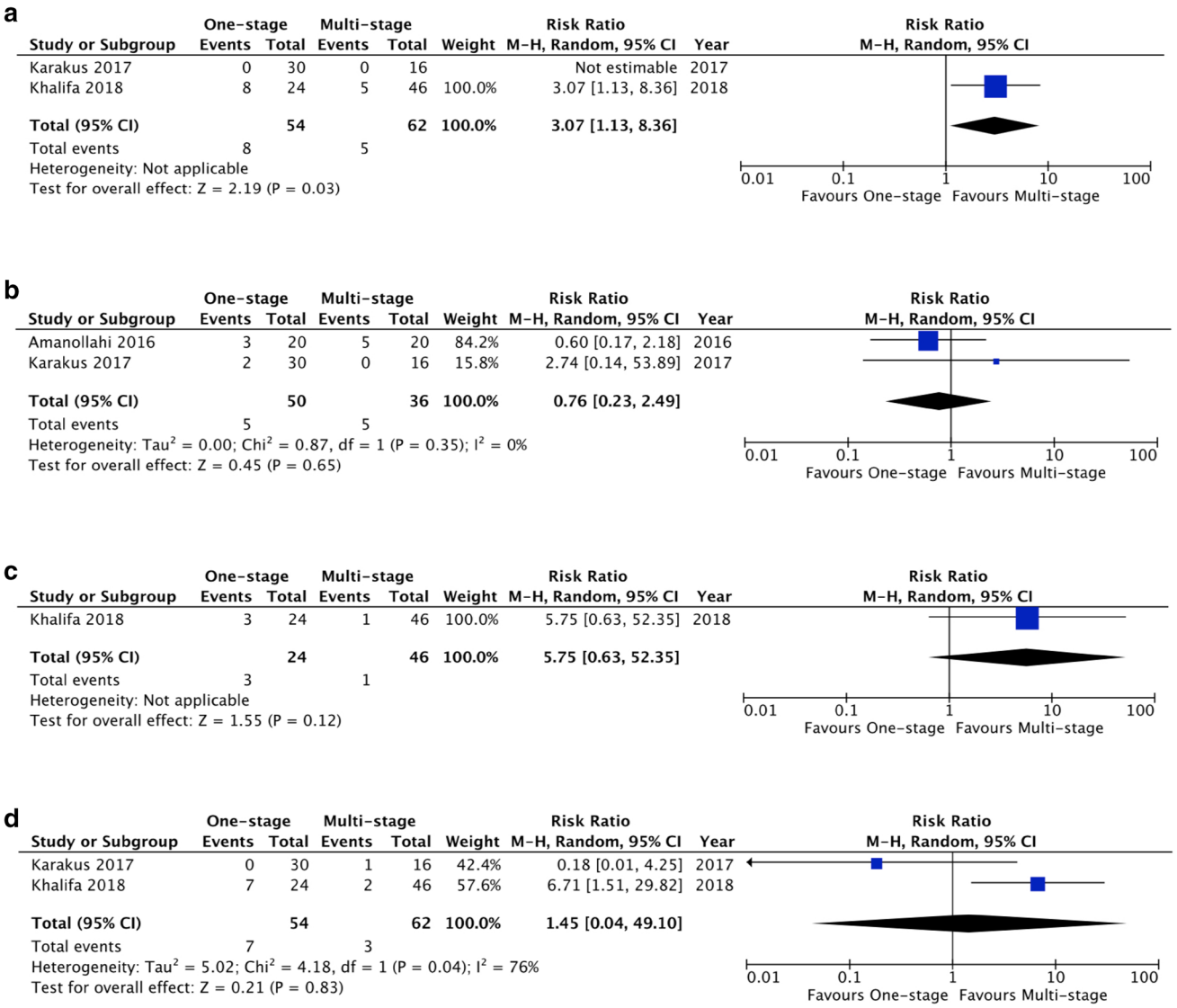

Fig. 3 **a** Comparison of the incidence of anorectal stenosis following one-stage repair vs. multi-stage approach; **b** comparison of the incidence of rectal prolapse; **c** comparison of the incidence of anal retraction; **d** comparison of the incidence of redo sagittal anorectoplasty

However, the present study demonstrates that one-stage SARP was associated with an increased risk of wound infection (24.3 ± 8.7%) compared to multi-stage approach (10.9 ± 2.5%; $p < 0.01$; Fig. 2a) and with an increased risk of wound dehiscence/disruption (16.2 ± 4.8% vs. 2.4 ± 1.1%; $p < 0.01$; Fig. 2b). A rescue colostomy was then necessary in 10.8 ± 7.3% patients in the one-stage approach group. As a possible consequence of the increased rate of wound complications following one-stage repair [2, 25], the incidence of anorectal stenosis was higher in this group of patients (14.8 ± 8.3%) compared to those who had multi-stage repair (8.1 ± 2.7%; $p < 0.05$; Fig. 3a).

With regards to others early complications, no differences were found with regards to rectal prolapse (10.0 ± 2.1% vs. 13.9 ± 6.2%, respectively; p = ns) and anal retraction (12.5% vs. 2.2%, respectively; p = ns). Similarly, the incidence of redo SARP was found to be similar between one-stage repair and multi-stage approach (12.9 ± 7.3% vs. 4.8 ± 0.8%; p = ns). However, it has been reported that the single-stage procedure requires redo SARP in up to 40% of cases, which questions the real advantages of one-stage repair [20, 30].

Patients with RVF usually have well-developed muscles and nerves. Therefore, RVF is associated with good prognosis with regard to fecal continence at long-term follow-up [21, 34]. Some series reported that approximately 90% of patients with corrected RVF would develop normal continence by the age of 3 years [22, 32]. Levitt and Peña reported that after RVF repair 90% of children had voluntary bowel movements and 36% had soiling by the age of 3 years [21]. In our meta-analysis, the prevalence of voluntary bowel

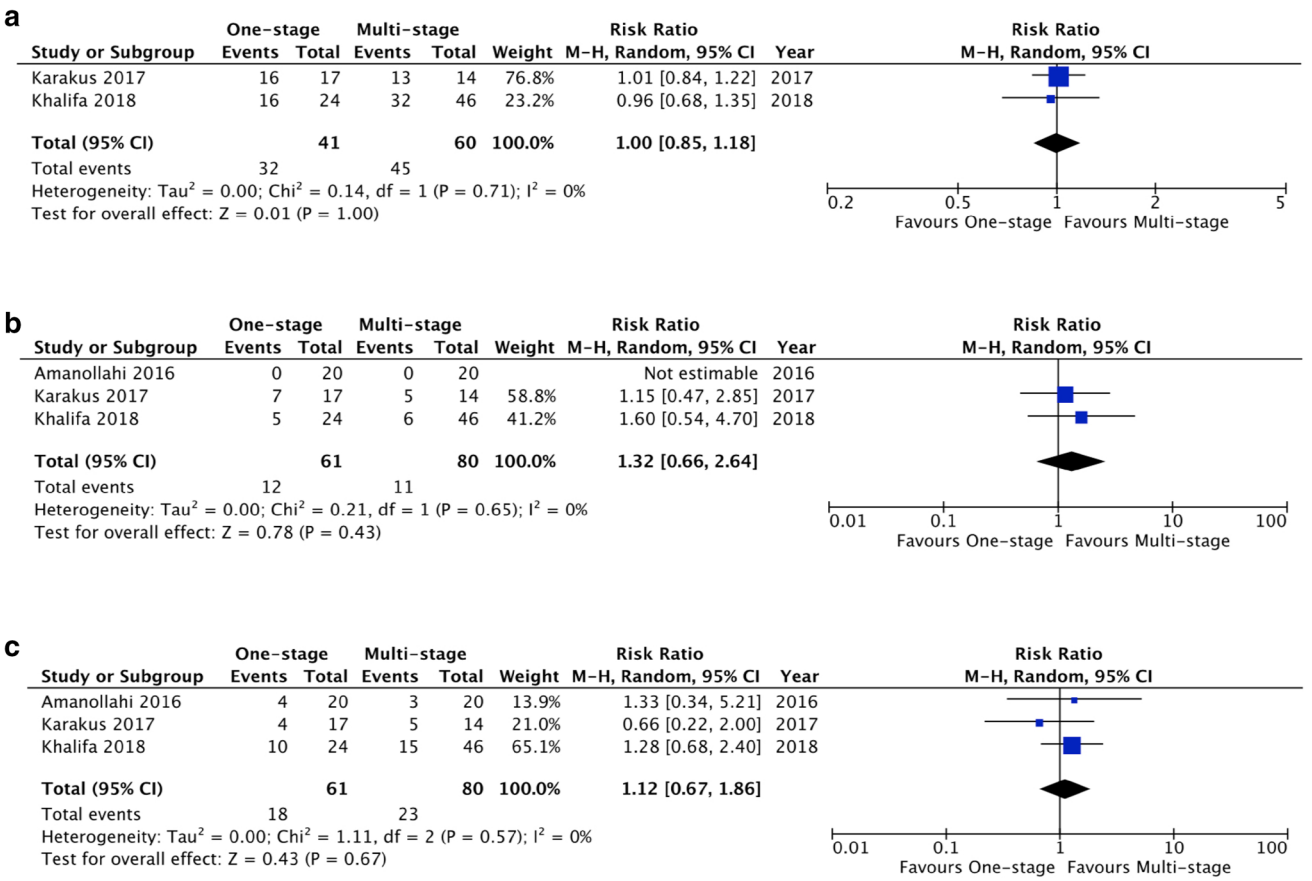

Fig. 4 **a** Comparison of the incidence of voluntary bowel movement between one-stage repair compared to multi-stage approach at long-term follow-up; **b** comparison of the incidence of soiling; **c** comparison of the incidence of constipation

movement did not differ in one-stage group (78.0 ± 6.8%) in comparison with staged SARP (75 ± 5.8%; p = ns; Fig. 4a). Similarly, the prevalence of soiling was similar after one-stage (19.7 ± 10.3%) and multi-stage repair (13.7 ± 8.9%; p = ns; Fig. 4b). It was estimated that 55% of patients with RVF would develop constipation [21]. However, in our study constipation did not differ between one-stage (29.5 ± 5.4%) and multi-stage repair (28.7 ± 4.4%; p = ns; Fig. 4c).

Limitations of the study

We acknowledge that there are some limitations in our present study, which are mainly due to the fact that a meta-analysis relies on the quality of the studies and data available in the literature. In our meta-analysis, two out of three studies included were retrospective and only one was a RCT (Table 2). None of the studies reached the gold standard cut-off on MINORS of 19.8 out of 24 (Table 4). Moreover, one paper reported the results of anterior SARP comparing one-stage vs. two-stage approach [12], whereas two studies included patients who undergone a posterior SARP comparing one-stage vs. three-stage approach [10, 11]. None of the papers provided sample size calculations, none of the studies reported a blinded evaluation of objective endpoints, and some of the follow-up periods were not appropriate to evaluate the continence of these patients (Table 3).

However, when independently assessed by two authors using AMSTAR, the present systematic reviews and meta-analysis received a relevant score (Supplementary file 2) and the PRISMA checklist was completed (Supplementary file 3).

Conclusion

In females with recto-vestibular fistula, the SARP performed without protective colostomy increases the risk of postoperative complications, such as wound infection, dehiscence, and anorectal stenosis. Nevertheless, the one-stage approach seems not to be associated with reduced fecal continence and constipation in comparison with the multi-stage repair. However, in the absence of prospective, randomized long-term studies evaluating the early and late results and

Table 4 Risk of bias assessment for individual studies using methodological index for non-randomized studies (MINORS) [13]

Item	Amanollahi et al. [18]	Karakus et al. [19]	Khalifa et al. [20]
1. A clearly stated aim	2	2	2
2. Inclusion of consecutive patients	2	2	2
3. Prospective collection of data	2	0	0
4. Endpoints appropriate to the aim of the study	2	2	2
5. Unbiased assessment of the study endpoint	0	0	0
6. Follow-up period appropriate to the aim of the study	1	2	2
7. Loss to follow-up less than 5%	0	1	2
8. Prospective calculation of the study size	0	0	0
9. An adequate control group	2	2	2
10. Contemporary groups	2	2	2
11. Baseline equivalence of groups	2	2	2
12. Adequate statistical analyses	2	2	2
Total score	17	17	18

0 not reported, *1* reported but inadequate, *2* reported and adequate

complications of one-stage repair of RVF, the decision of performing a protective colostomy or primary repair without colostomy depends on the experience of the surgeon and the general conditions of the patient.

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Compliance with ethical standards

Conflict of interest Authors have no potential conflicts of interest for this study.

Ethical approval Not applicable, since the study was a systematic review and meta-analysis.

Informed consent Not applicable, since the study was a systematic review and meta-analysis.

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