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The outcome of Bishop-Koop procedure compared to divided stoma in neonates with meconium ileus, congenital intestinal atresia and necrotizing enterocolitis

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Abstract

To determine the potential value and suitability of Bishop-Koop procedure (BK) compared to divided stoma (DS) in neonates with meconium ileus (MI), congenital intestinal atresia (CIA), and necrotizing enterocolitis (NEC).

A retrospective data collection from 2000 to 2019 on neonates undergoing BK and DS formation and closure for MI, CIA, and NEC was conducted. Ostomy related complications following both procedures were analyzed.

One hundred two consecutive patients managed with a BK (n = 57, 55.8%) and DS (n = 45, 44.2%) for MI (n = 38, 37.2%), CIA (n = 31, 30.5%), and NEC (n = 33, 32.3%) were analyzed. Mean operating time for ostomy creation did not differ significantly between BK and DS groups (156 ± 54 vs 135 ± 66.8 min, $P = .08$). The prevalence of stoma-related complications following BK and DS formation was 8.7% and 31.1%, respectively ($P = .005$). The complication rate after BK and DS closure was 3.5% and 6.7%, respectively ($P = .65$). The operating time for ostomy reversal and length of hospital stay after stoma closure were significantly shorter in BK group (82.2 ± 51.4 vs 183 ± 84.5 min and 5.5 ± 2.7 vs 11.3 ± 3.9 days, $P < .001$).

BK procedure is safe, reliable, and suitable technique in neonatal surgery with low complications rate following ostomy creation as well as shorter operating time and length of hospital stay after ostomy closure compared to DS ostomies. Surgeons should keep this technique as an alternative approach in their repertoire.

Abbreviations: BK = Bishop-Koop procedure, CIA = congenital intestinal atresia, DS = divided stoma, MI = meconium ileus, NEC = necrotizing enterocolitis, OIC = ostomy in continuity, PA = primary anastomosis, SE = Santulli enterostomy.

Keywords: Bishop-Koop procedure, complications, congenital intestinal atresia, divided stoma

1. Introduction

A variety of neonatal intestinal disorders, such as meconium ileus (MI), congenital intestinal atresia (CIA) or necrotizing enterocolitis (NEC), requires urgent surgical intervention. The different operative strategies include creation of a primary anastomosis (PA) following bowel resection, formation of a temporary diverting stoma (DS), or an ostomy in continuity (OIC) such as Bishop-Koop (BK) or Santulli enterostomy (SE). The intraoperative decision-making process is based on individual patient conditions, severity of underlying disease, training backgrounds and personally expertise of the surgeon.

The PA following bowel resection promotes a single-stage procedure that maintains intestinal autonomy. However, without creation of a proximal protective enterostomy, the incidence of anastomotic leaks ranges between 1.6% and 31%.[1–7] Conversely, a 2-stage restoration of intestinal continuity using formation of the diverting enterostomy has been advocated for neonates who are at increased risk of anastomotic complications due to peritonitis (NEC) or size discrepancy between the dilated proximal and the unused distal bowel (CIA, MI). Even so, stoma related complications and the need for a second operation to close the stoma are issues of concern. The frequency of ostomy related complications ranges from 18% to 42% following ostomy formation[8–10] and is as high as 70% following ostomy closure.[11–13] Although the decision between PA or DS in the treatment of MI, CIA, and NEC remains an area of controversy, both treatment strategies are accepted as standard surgical modalities.

An alternative approach to PA and divided stoma (DS) is OIC, including Bishop-Koop procedure (BK) and Santuli enterostomy, which both allow sufficient bowel decompression while also preserving intestinal transit. Following resection of the intestinal segment, the end of the proximal limb is anastomosed to the side of the distal (BK) limb and the end of the distal limb is brought out as the enterostomy (also known as the "chimney"). The mirror image, in which the end of the proximal limb is used for the enterostomy, represents the SE.[14]

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Though the BK was initially designed for the treatment of infants with MI, it has been gradually applied in neonates with NEC[15,16] and CIA.[17–19] Recently, BK has also been utilized in the management of children with short bowel syndrome[20] and severe jejunoileal atresia.[21]

In our practice, BK has been used as a standard treatment for MI, CIA, and NEC, in addition to PA and DS. We have utilized this technique since 2000 with the intention of diverting the fecal stream and protecting the anastomosis. The preservation of intestinal continuity minimizes intestinal fluid loss seen in DSs. In cases of intestinal transport difficulty, the "chimney" acts as a safety vent, partially decompressing the anastomosis and reducing the risk of anastomotic breakdown. Furthermore, the "chimney" can also be used as an access point for diagnostic endoscopic procedures or contrast enema.

Despite its historical significance, the BK has largely fallen out of use in pediatric surgical practice as many pediatric surgeons prefer PA when clinically appropriate. We have found a paucity of comparative information in the recent literature exploring the suitability and safety of BK in neonatal surgery. Therefore, we decided to analyze and report our data to determine the clinical impact and suitability of BK procedure compared to DS in neonates with MI, CIA, and NEC.

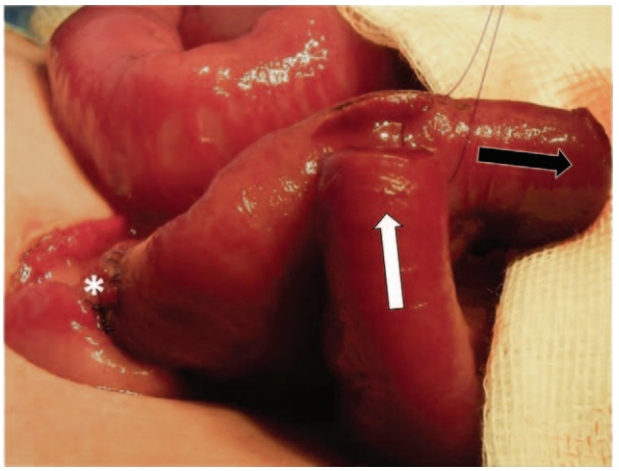

Figure 1. Intraoperative photograph of patient with atresia of the terminal ileum and non-used microcolon (not shown) shows primary anastomosis (asterisk) and protective Bishop-Koop enterostomy following resection of the atretic bowel segment. The end of the proximal intestine (white arrow) is anastomosed to the side of the distal intestine. "Chimney" segment (black arrow) will be exteriorized through a small incision.

2. Methods

2.1. Study design and patient selection

All patients who underwent BK and DS formation and closure from January 2000 to January 2019 at the Medical Center – University of Freiburg, Center for Surgery, Department of Pediatric Surgery were included in this retrospective case-control study. Data collection included gestational age, birth weight, age and weight at time of operation, indications for surgical therapy, level of intestinal diversion, time to stoma function, time to stoma closure and complications directly related to the ostomy. The institutional review board of the University of Freiburg granted approval for this retrospective study (#96/15).

2.2. Exclusion criteria

Children older than 6 months of age and neonates with associated congenital anomalies, such as anorectal malformation or Hirschsprung disease, were excluded from the study. Additionally, neonates with congenital syndromes affecting multiple organ systems were excluded. Children with CIA, MI, or NEC were also excluded from the study if the medical record was incomplete.

2.3. Surgical technique

2.3.1. Bishop-Koop procedure. The formation of Bishop-Koop anastomosis took place in the operation theater under general anesthesia. The procedures were performed by 1 of 4 experienced pediatric surgeons. After abdominal exploration, segmental bowel resection or other appropriate surgical intervention to solve the causative problem was performed. For reconstruction, the distal bowel was fashioned into a stoma and anastomosed side-to-end into the proximal bowel (Fig. 1). We used continuous single layer suture technique. The extension of the distal bowel was exteriorized as an end enterostomy or "chimney." A postoperative drainage was not routinely used.

2.3.2. DS procedure. A DS was defined as bowel discontinuity, regardless of the distance between the proximal and the distal ends of the stoma. All divided ostomies were fixed to fascia.

2.4. Definitions of underlying medical conditions

MI was defined as intestinal obstruction characterized by the impaction of thick, inspissated, protein-rich, adhesive and dessicated meconium in children with or without cystic fibrosis (meconium plug syndrome). NEC was defined as ischemic necrosis of the intestinal mucosa with pneumoperitoneum secondary to intestinal perforation leading to resection of necrotic bowel segment. Infants with spontaneous intestinal perforation were included in the NEC group. CIA was defined as neonatal intestinal small bowel obstruction caused by jejunal or ileal atresia with large diameter ratio of the proximal and distal lumens.

2.5. Complications following ostomy formation and closure

We analyzed all ostomy related complications following formation and closure, including the following: parastomal hernias, mucocutaneous separation, necrosis, prolapse, retraction, stenosis, peristomal skin irritation, peristomal infection abscess, and fistula, as well as acute bowel obstruction for both BK and DS procedures.

2.6. Statistics

Results are presented as absolute numbers, a percentage (%) frequency, a mean with corresponding standard deviation or a median with range when appropriate. Statistical analysis was performed using SPSS Version 23. The Student *t* test was used to determine statistical significance for data with normal distribution, and the Mann–Whitney *U* test was used to compare data that were not normally distributed. The chi-square and Fisher exact tests were used to compare complication rates between the

Table 1

Demographic and clinical patient characteristics of the BK and DS group.

	Bishop-Koop Anastomosis n = 57	DS n = 45	*P value*
Gestation age [d, range]	29.6 ± 8.1 (21–42)	30.5 ± 5.9 (23–31)	.62
Birth weight [g; range]	1800 ± 1239 (340–5800)	1580 ± 1051 (490–3625)	.34
Sex [N;%]			.84
Male	33 (57.9%)	25 (55.6%)	
Female	24 (42.1%)	20 (44.4%)	
Diagnosis [N;%]			.15
NEC/FIP	14 (24.6%)	19 (42.2%)	
MI	23 (40.3%)	15 (33.3%)	
CIA	20 (35.1%)	11 (24.5%)	
Ostomy creation			
Age [d, range]	25.6 ± 34.3 (1–157)	21.7 ± 25.7 (1–88)	.52
Weight [g; range]	2272 ± 1789 (550–9245)	1983 ± 1048 (500–1048)	.33
Operating time [min; range]	156 ± 54 (79–287)	135 ± 66.8 (82–324)	.08
Revisional surgery [N;%]	5 (8.7%)	14 (31.1%)	.005
Ostomy closure			
Age [d; range]	243.3 ± 174.7 (29–807)	190.5 ± 86.2 (79–380)	.06
Weight [g; range]	5468 ± 2399 (1170–10000)	4630 ± 2081 (1780–9245)	.06
Operating time [min; range]	82.2 ± 51.4 (44–110)	183 ± 84.5 (49–226)	<.001
Length of hospital stay [d; range]	5.5 ± 2.7 (2–17)	11.3 ± 3.9 (6–23)	<.001
Complications [N;%]	2 (3.5%)	3 (6.7%)	.65

CIA = congenital intestinal atresia, FIP = focal intestinal perforation, MI = meconium ileus, NEC = necrotizing enterocolitis.

groups. A significance level of $< .05$ was adopted. All P values were 2-sided.

3. Results

Over the 19 year study period, 57 (55.8%) BK and 45 (44.2%) DS procedures were created and all of them subsequently closed at our institution. There were no intraoperative complications or hospital mortality in both groups. Demographic data and clinical characteristics are summarized in Table 1. There were no statistically significant differences regarding clinical characteristics between patients who had a BK and those who had DS. The proximal segment of BK and DS was localized in the jejunum (n = 12, 21.0% vs 17, 37.7%), ileum (n = 42, 73.6% vs n = 26, 57.7%) or large intestine (n = 3, 5.4% vs n = 2, 4.6%). The distal vent of BK and DS was localized in the jejunum (n = 12, 21.0% vs n = 15, 33.3%), the ileum (n = 41, 72.0% vs n = 23, 51.2%) or the large intestine (n = 4, 7.0% vs n = 7, 15.5%). The mean duration of stoma use was 246.1 ± 82 days in BK group and 221.5 ± 93 days in DS group ($P = .15$). In 2 patients (3.5%), the BK stoma closed spontaneously. Nevertheless, these patients also underwent formal ostomy closure to restore the integrity of the abdominal wall.

Complications leading to revisional surgery following ostomy formation occurred in 19 (18.6%) patients. Patients with DS developed significantly more stoma related complications compared to patients in BK group (14 (31.1%) vs 5 (8.7%), $P = .005$). Complications in children with DS included prolapse of stoma (n = 7, 50%), stoma stenosis (n = 5, 35.7%) and parastomal hernia (n = 2, 14.3%). Among the patients with BK procedure, small bowel obstruction without necrosis due to the rotation of bowel around the ostomy vent was seen in 4 patients and stoma prolapse in 1 patient. The clinical characteristics of patients with complications following BK formation are shown in Table 2. There was no evidence of anastomotic leakage or stricture.

To examine the influence of underlying diagnosis, gestational age, birth weight, age and weight at time of ostomy creation, and duration of ostomy use on surgical complications, additional subanalysis was performed. The data is summarized in Table 3. The differences among patients with and without ostomy related complications within BKP and DS groups were not statistically significant.

Rates of complications following ostomy closure were generally low in both groups (n = 2, 3.5% vs n = 3, 6.7%, $P = .65$). Wound infections were seen in 2 (3.5%) patients within the BK group. Complications among DS group included

Table 2

Complications following BK formation.

Patient	Sex	Diagnosis	Gestation Age [d]	Birth weight [g]	Age at ostomy formation [d]	Weight at ostomy formation [g]	Time to complications [d]	Revisional surgery
1	f	MI	41.1	3625	1	3625	4	BK
2	f	NEC	28.0	595	24	1020	15	BK
3	m	NEC	26.2	980	84	2460	1	BK
4	m	MI	29.4	1900	1	1900	242	BK
5	m	CIA	32.7	3650	52	4800	189	Closure

BK = Bishop-Koop procedure.

Table 3

Outcomes between BK and DS.

	BK (n = 57)		DS (n = 45)		
	Complications (n = 5)	No complications (n = 52)	Complications (n = 14)	No complications (n = 31)	*P value*
Diagnosis					
MI	2	21	7	12	.23
CIA	1	19	2	13	
NEC/FIP	2	12	5	6	
Gestation age [d, range]	31.5 ± 5.8 (26.2 – 41.1)	29.2 ± 3.6 (25.6–42.0)	31.1 ± 6.2 (25–3)	29.3 ± 5.9 (24–31)	.34
Birth weight [g; range]	2150 ± 1438 (595–3650)	1980 ± 1141 (540 – 5800)	1280 ± 1251 (490 – 3450)	1452 ± 910 (670–3625)	.11
Age at operation [d, range]	32.4 ± 35.7 (1–84)	25.8 ± 32.3 (1–157)	22.5 ± 24.2 (1–77)	23.3 ± 20.0 (1–88)	.23
Weight at operation [g; range]	2761 ± 1480 (1020–4800)	2357 ± 1980 (550–9245)	2310 ± 1030 (720–1048)	2310 ± 1030 (500–1020)	.31
Duration of stoma use [w, range]	81.8 ± 92.6 (8 – 230)	231.1 ± 79.2 (152–311)	92.2 ± 73.1 (24–467)	210 ± 43.1 (24–467)	.22

BK = Bishop-Koop procedure, CIA = congenital intestinal atresia, DS = divided stoma, FIP = focal intestinal perforation, MI = meconium ileus, NEC = necrotizing enterocolitis.

anastomotic leakage (n = 1, 33.3%), anastomotic stricture (n = 1, 33.3%), and wound infection (n = 1, 33.3%). Wound infection was managed conservatively. Other complications required revisional surgery.

The operating time for BK closure was significantly shorter compared to DS (82.2 ± 51.4 vs 183 ± 84.5 min, $P < .001$) (Fig. 2). Patients with BK exhibited rapid recovery and the length of hospital stay was significantly shorter compared to the DS group (5.5 ± 2.7 vs 11.3 ± 3.9 days, $P < .001$) (Fig. 3).

4. Discussion

To circumvent the surgical problems associated with PA if created without protective ostomy as well as stoma-related morbidity, we utilized the BK technique as a standard procedure to treat neonates with MI, CIA, and NEC.

The present study demonstrated a lower complications rate after BK creation compared with DS. BK closure was also associated with reduced operating time and shorter length of hospital stay. However, in spite of its potential benefits, data on BK utilization in neonates are very scarce. Table 4 summarizes the available literature on use of BK in neonates.

In our study, the frequency of complications following BK formation was 8.7% and 3.5% following its closure. The range of BK related complications reported in the literature varies from 0.0% to 19.9% for BK ostomy creation and from 0.0% to 7.4% for its closure.[17,22,23] The most frequent complication was the rotation of bowel around the ostomy vent, occurring in four patients in our series (7.0%). This unique type of complication can be regarded as specific to BK enterostomy and has also been reported with low incidence in other studies.[17] We also observed 1 case of Bishop-Koop stoma prolapse. This type of stoma related complication is reported to be a very rare event.[16] No cases of stoma necrosis, retraction, parastomal hernia, peristomal skin breakdown or wound cellulitis were observed. Furthermore, anastomotic leakage or stenosis did not occur in our BK patient cohort. According to the literature, anastomotic leakage following BK formation occurs in up to 5% of patients.[17,20,21] In contrast, prior studies demonstrate a variable rate of anastomotic complications following PA if created without protective ostomy ranging from 15% to 30% for MI,[1,2,24,25] from 6% to 10% for CIA,[26–28] and from 5% to 12% for NEC.[5,7,10,29] We hypothesize that the lower rate of anastomotic complications in BK compared to PA is due to presence of a diverting ostomy, which serves as a vent and thereby protects the anastomosis.

Some may argue that the BK procedure is time-consuming because it requires an anastomosis. However, in the current study, the operating time for ostomy formation did not significantly differ between BK and DS groups. Furthermore, the premature neonates with NEC and SIP represent a particular patient group in our study cohort. Due to immature motility of

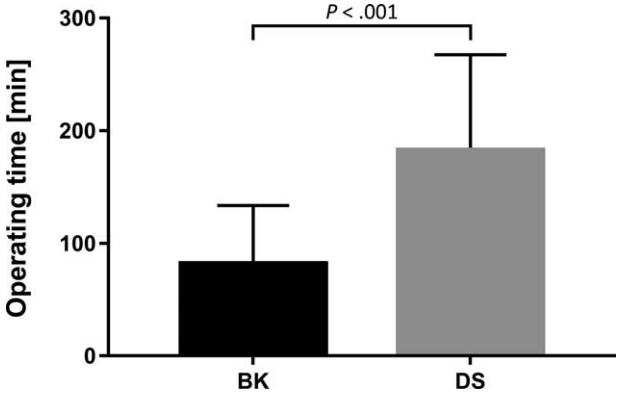

Figure 2. Means and standard deviations of the operating times for ostomy closure for BK and DS. BK = Bishop-Koop procedure, DS = divided stoma.

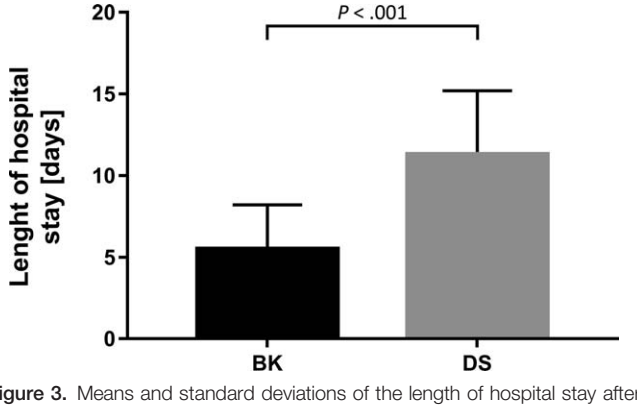

Figure 3. Means and standard deviations of the length of hospital stay after ostomy closure for BK and DS. BK = Bishop-Koop procedure, DS = divided stoma.

Table 4

Summary of BK literature.

Nr.	Author	Year	No. of procedures	Diagnosis	The rate of complications following BK creation n (%)	The rate of complications following BK closure n (%)
1	Bishop[14]	1957	1	MI/CF	0 (0)	–
2	Dickson[30]	1970	4	CIA (DA/JA)	0 (0)	–
3	McPrtlin[31]	1972	–	MI	–	–
4	Valerio[32]	1978	4	LBE	–	–
5	Caniano[33]	1987	7	MI	–	–
6	Palmieri[34]	1993	1	AMI/SI	0 (0)	0 (0)
7	Herzog[35]	1996	1	BLT	–	–
8	Murshed[36]	1997	2	MI	0 (0)	–
9	Fleet[18]	2000	1	CIA	0 (0)	0 (0)
10	Wit[17]	2000	27	MI, NEC, CIA	3 (11.1%)	2 (7.4%)
11	Kumaran[28]	2002	6	CM	–	–
12	Arsalani-Zadeh[37]	2010	2	AMI	0 (0)	0 (0)
13	Mirza[16]	2010	1	SI	–	–
14	Burjonrappa[38]	2011	8	CIA (JA/CA)	1 (12.5%)	0 (0)
15	Haxhija[19]	2011	3	CIA (CA)	0 (0)	0 (0)
16	Kayastha[39]	2011	1	CIA	0 (0)	0 (0)
17	Watanabe[40]	2011	3	ELSCH/SI	0 (0)	0 (0)
18	Boczar[41]	2015	8	MI	0 (0)	–
19	Hasan[22]	2017	21	MI	4 (19.0%)	–
20	Sehgal S[20]	2018	12	CBS	0 (0.0%)	1 (8.3%)
21	Peng Y[21]	2018	41	SJIA	4, 9.7%	–
22	Present study	2019	57	MI, NEC, CIA	5 (8.7%)	2 (3.5%)

AMI = acute mesenteric ischaemia in adults, BLT = Bowel-liver transplantation in an infant with microvillous inclusion disease, CA = colonic atresia, CDS = short bowel syndrome, CM = congenital malformations, DA = duodenal atresia, ELSCH = excessively long segment congenital hypoganglionosis, IA = ileal atresia, JA = jejunal atresia, LBE = Large bowel emergencies: emergency operation for carcinoma, diverticular disease or colitis in adults, SI = second intervention, SJIA = severe jejunoileal atresia (complex meconium peritonitis, type IIIb and type IV).

the bowel and peritonitis, the creation of DS seems to be safer than BKP. Nevertheless, all patients with NEC in the BK group survived and underwent successive ostomy closure. Moreover, we additionally compared operating times in the subgroup of patients with NEC and did not observed any statistical significant differences between BK and DS groups (150 ± 56.4 vs 130.2 ± 56.2, $P = .35$).

In the present study, only 2 patients in the BK group suffered from wound infections after ostomy reversal (n = 2, 3.5%); both were treated conservatively. This finding is in line with other studies in which the occurrence of wound infections after stoma closure reportedly range from 0.0% to 7.4%.[17,38] Conversely, in the DO group, more severe complications occurred following ostomy reversal, including anastomotic leakage and stenosis (n = 2, 4.4%) requiring revisional surgery. The differences are explained by the fact that there is no need for the creation of an anastomosis during BK takedown, with the reversal procedure consisting simply of chimney closure.

In our study, time to ostomy closure (246.1 ± 82 days in BK group and 221.5 ± 93 days in DS group) was found to be much longer than previously observed in other studies, in which mean time to reversal ranged from 84 to 177 days.[9,42–45] This discrepancy can be explained in part by surgeon's preference and our local protocol, but is not evidence-based. Surgical aspects that favor later ostomy closure include fewer postoperative abdominal adhesions,[46–48] anesthesiological aspects include potential adverse neurodevelopmental outcomes as a result of general anesthesia at a very young age.[49,50] Moreover, the unique anatomical advantage of BK anastomosis, which preserves intestinal continuity and maintains the fluid and electrolyte balance, relativizes the potential advantages of early ostomy closure. Nevertheless, there is currently no consensus for the optimal timing of ostomy reversal. In fact, although a number of studies have sought to determine the optimal time to close an enterostomy, there is no significant difference in the complication rate of infants whose enterostomy was reversed within 8 weeks (27–31%) or after 8 weeks (19–23%) following its creation.[11,51,52]

We are aware of several limitations of this study. These include a small patient population and the retrospective study design. Nevertheless, our study represents one of the largest series of BK procedures performed in neonates with gastrointestinal tract disorders.

In conclusion, Bishop-Koop procedure has a low rate of complications following its formation and a satisfactory operating time. Moreover, BK closure is associated with shorter operating time and shorter length of hospital stay compared to DS. Although it remains the responsibility of the surgeon to determine the utilization of treatment techniques, surgeons should keep this technique as an alternative approach in their repertoire. Nevertheless, without a sufficiently powered randomized controlled trial, no argument can be made regarding the superiority of one operative strategy over another.

Author contributions

Conceptualization: Illya Martynov, Jochen Raedecke, Joachim Schoenberger.

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