A New Appendicostomy Technique to Prevent Stomal Stenosis

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Abbreviations and Acronyms

ACE = antegrade continence enema

BMI = body mass index

Accepted for publication December 10, 2019.

No direct or indirect commercial, personal, academic, political, religious or ethical incentive is associated with publishing this article.

Study received institutional review board approval (IRB No. 591176).

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Purpose: Stomal stenosis has been reported to occur in 12% to 45% of patients following Malone antegrade continence enema and Mitrofanoff appendicostomy. The standard stoma technique entails excision of the distal appendix. We evaluated a novel technique with preservation of the appendiceal tip and vessels, and opening the lumen in a more proximal and vascular area to determine whether the incidence of stenosis would be decreased.

Materials and Methods: Medical records of patients who underwent appendicostomy for Malone antegrade continence enema or urinary diversion were retrospectively evaluated. We included cases with a minimum of 1 year of followup and those in which the distal portion of a complete appendix was oriented for use as the stomal end in the umbilicus. Variables such as age, gender, body mass index, antegrade continence enema or urinary diversion, open or laparoscopic approach, cecal and appendiceal adhesions, retrocecal position, cecal imbrication, technique and stenosis were recorded. Cox proportional hazards analyses were performed to determine association of covariates.

Results: A total of 123 patients met inclusion criteria. The incidence of stenosis following standard stoma technique was 13% (12 of 93 patients) with a median followup of 9.4 years. Of these cases 75% occurred within 1 year of surgery. Stomal stenosis did not occur after the new stoma technique in 30 patients with a median followup of 3.3 years. Only technique cohort (standard vs new) was associated with stenosis ($p=0.04$).

Conclusions: Stomal stenosis of appendicostomy may be lessened by preservation of the distal appendiceal vasculature and tip, and opening the lumen in a more proximal location.

Key Words: surgical stomas, enema, fecal incontinence, therapeutic irrigation, spinal dysraphism

ALTHOUGH considered vestigial, the vermiform appendix is a vital structure for urological surgeons and their patients. The appendix typically has adequate length, lumen size and mobile vasculature to serve as a conduit. Thanks to pioneers in the field, notably Mitrofanoff[1] and Malone et al,[2] we have been able to use the appendix for continent urinary diversion and delivery of antegrade continence enemas.

The standard technique for creation of an umbilical appendicostomy is excision of the appendiceal tip, antimesenteric spatulation and inversion of an inferior triangular skin flap into the spatulation. The superior edges of the appendiceal opening are sewn to the upper edge of umbilical skin. The most common complication after appendicostomy is stomal stenosis, which is reported to occur in 12% to 45% of patients.[3–12] A testament to the high incidence of stenosis is the preponderance of stoma techniques. Most of these are directed to nonumbilical

0022-5347/20/2036-1200/0
THE JOURNAL OF UROLOGY®

https://doi.org/10.1097/JU.0000000000000711
Vol. 203, 1200-1206, June 2020
Printed in U.S.A.

stomas with creation of skin flaps that drop below the skin surface such that the mucosa is hidden.[13,14]

For umbilical stomas the inferior V-flap is generally soft, mobile and well perfused, similar to skin used in nonumbilical stomas. On the other hand, the posterior umbilical skin is usually hard and tough. In 2000 the author found that complete excision of posterior umbilical skin allows an easier anastomosis without compromising the appearance of the hidden stoma. With the goal to lessen stenosis the author in 2012 stopped resecting the end of the appendix and used the principle of a loop ileostomy and ureterostomy, which ensures better vascularization.[15] Unlike loop ileostomy, a knuckle of bowel is not formed. The distal end of the appendix is kept above the fascia with preservation of the appendiceal tip and vessels and creation of the stoma more proximally.

In addition to stoma technique, there are other patient, anatomical and surgical variables that may be associated with stenosis, such as age, obesity, extensive mesenteric mobilization, "de-hinging" a twisted appendix and cecal imbrication. The goal of this retrospective study was to determine if any of these factors or appendicostomy technique was associated with stenosis.

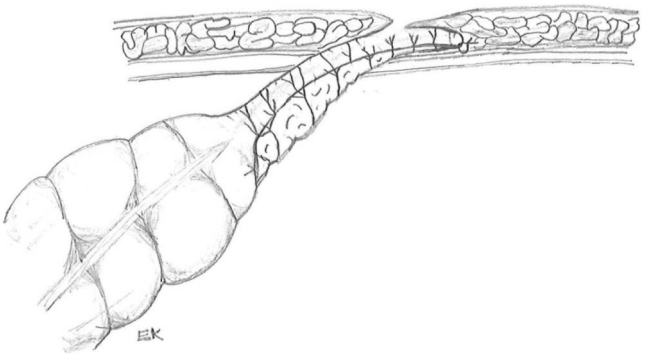

Figure 1. Lateral view of new appendicostomy shows tip of appendix secured to external oblique fascia superior to umbilicus and antimesenteric stoma incision on appendix at inferior aspect of umbilicus.

METHODS

Beginning in January 2000 standard appendiceal stoma formation was performed by fashioning a V-flap from the inferior umbilical skin, resection of the posterior umbilical skin, delivery of the appendix through a fascial incision just inferior to the umbilicus, ligation of the appendiceal artery at the desired length, excision of the distal appendix, antimesenteric spatulation and then maturation to the inverted flap of umbilical skin with interrupted 5-zero polyglactin suture. A catheter was left in place for 4 weeks, followed by institution of intermittent catheterization.

The new appendicostomy technique was initiated in 2012 (fig. 1). After fashioning a triangular skin flap and excising the posterior skin as described the superior umbilical skin is retracted anteriorly. By gentle spreading under the fat, the fascia is exposed and a small space is created superior to the umbilicus. A 5-zero polyglactin suture is placed in the fascia at the top of the space. The appendix is brought through the fascial incision inferior to the umbilicus and the tip is secured to the fascia above the umbilicus (fig. 2). A sagittal incision is made on the anterior antimesenteric appendix, opening the lumen from mid umbilicus to below the umbilicus. The skin flap is sewn into the lower aspect of the opening. The lateral and superior edges of the appendiceal opening are sewn to the pliable undermined umbilical skin edges. Detailed images of this technique can be found at https://health.ucdavis.edu/urology/specialties/pediatric_urology/handouts-physicians-supplements-journal-manuscripts.html.

The stoma technique was not changed based on surgical approach (open vs laparoscopic). For open cases the cecum (for ACE) or bladder (for urinary diversion) was secured to the anterior abdominal wall after stoma completion.

After institutional review board approval (IRB No. 591176) medical records of patients who underwent appendicostomy for Malone ACE or urinary diversion between 2000 and 2018 were evaluated. Only cases with a minimum of 1 year of followup and those in which the distal portion of a complete appendix was oriented for use as the stomal end in the umbilicus were included. Patients

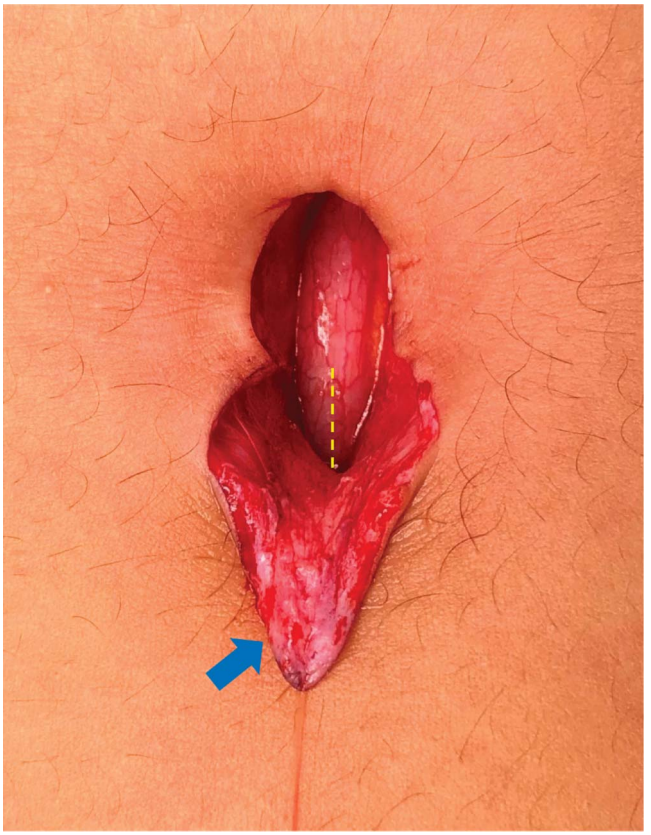

Figure 2. Distal appendix lying in umbilical bed. Arrow indicates inferior umbilical skin flap. Dotted line indicates antimesenteric incision on appendix for stoma.

who had a nonumbilical stoma were excluded to isolate the impact of the technical modification, which was limited to umbilical stomas. Patients with split appendix or cecal extension technique were also excluded.

Variables that were recorded included date of surgery, age at surgery, gender, BMI, ACE or urinary diversion, surgical approach (laparoscopic vs open), appendiceal position (retrocecal or other), need to excise appendiceal adhesions, cecal imbrication, stoma location, stoma technique and length of followup. BMI and percentiles were calculated using the Centers for Disease Control and Prevention calculator (https://www.cdc.gov). Patients were categorized as normal, overweight (85th to 94th percentile) or obese (95th to 99th percentile). Stomal stenosis was defined as difficulty placing a catheter, which necessitated either long-term catheter (stopper) use and/or revision. Dates of first occurrence of stenosis and revision were recorded.

Patient and surgery characteristics were compared using Wilcoxon rank sum tests for continuous variables and Fisher exact test for categorical variables. Time to stenosis was modeled by patient and surgery characteristics using Cox proportional hazards models. Models were fitted using Firth bias reduced maximum likelihood [1], as some variable levels had no stenoses. Analyses were conducted using R 3.6.1 software (R Project for Statistical Computing, http://www.r-project.org). Firth bias reduced maximum likelihood was fitted using the R package, coxphf version 1.13.

RESULTS

Over the 19-year period 123 patients (93 with standard stoma, 30 with new stoma) met inclusion criteria, of whom 113 had neuropathic disease. Four patients had a history of posterior urethral valves, 2 had prune belly syndrome, 2 had bilateral ectopic ureters with outlet dysfunction and 2 had prostatic rhabdomyosarcoma.

After initial laparoscopic ACE surgery the appendix was repurposed in 2 patients to an appendicovesicostomy at 5 and 10 years. Two patients with a standard stoma stopped using the ACE at 5 and 8 years, and the followup periods were recorded using those dates. Six patients, all with a standard stoma, were excluded. Two patients moved within a year of surgery. One patient was noncompliant within 6 months of surgery. Two patients were excluded due to difficult catheterization proximal to the stoma, 1 due to appendicocecal angulation and 1 due to a "crunchy" appendiceal lumen found during surgery that persisted after the catheter was removed.

Patient, anatomical and surgical variables are detailed in table 1. All data points were available except height measurements in 7 patients. Among the variables there was no significant difference between the cohorts. At surgery 52% of patients were overweight or obese. Obesity was more prevalent in the new stoma patients, although the difference was not statistically significant.

The incidence of stenosis after standard stoma surgery was 13% (12 of 93 patients) with a median followup of 9.4 years. Of these cases 67% occurred within 6 months and 75% within 1 year of surgery (Kaplan-Meier curves, fig. 3). Some patients with stenosis used a stopper or indwelling catheter until corrective surgery. Revision surgeries were performed 2 to 13 months after first occurrence and all patients but 1 have been free of stenosis for 4 to 17 years following revision.

After the new stoma technique no patient had stenosis during a median followup of 3.3 years. Although stenosis was defined as difficulty placing a catheter that necessitated either long-term catheter (or stopper) use and/or revision, no patient has required dilation or steroids.

Cox proportional hazards models showed no association of stenosis with patient, anatomical or surgical variables except stoma technique ($p=0.04$, table 2). Patients with a new stoma had an eightfold lower hazard of stenosis than those with a standard stoma (HR 0.125, 95% CI 0.00–0.95).

There has been no morbidity from the preservation of the distal tip, nor can it be palpated or appreciated on physical examination. Although not objectively analyzed, the author has not found any difference in appearance of the umbilical stomas, which are nearly impossible to visualize without probing the deep umbilicus.

DISCUSSION

In 1980 Mitrofanoff described use of the appendix for urinary diversion in 16 patients.[1] The distal tip of the appendix was excised and implanted into the bladder with the wider cecal end preserved for a stoma. Ten years later Malone et al described appendicostomy for fecal dysfunction with detachment and reversal of the appendix and placement into a cecal tunnel.[2] The stoma was created in the right lower quadrant by fashioning a skin tube sewn to the cecal cuff. Subsequently Griffiths and Malone authored a case series (21 subjects) describing the Malone antegrade continence enema, enshrining the eponym.[16] Modifications included leaving the appendix in situ with excision of the tip and spatulation. Unfortunately more than half the patients had stomal stenosis or breakdown, presaging the outcomes that we see today. Contemporary large series demonstrate stenosis in 12% to 45% of patients.[3–12,17,18] Some institutions have recommended leaving a "stopper" in place for 6 months, or indefinitely.[19]

In this analysis we tried to evaluate every known variable that has been associated with stenosis. Yet the most important variable is time. Most long-term

Table 1. *Patient and surgery characteristics by stoma technique*

	Standard Stoma	New Stoma	Overall	p Value
No. pts	103	30	133	
Age at surgery:				0.08 (Wilcoxon rank-sum test)
Median yrs (range)	7.8 (4–25)	10.2 (5–23)	8.1 (4–25)	
No. pts	93	30	123	
Median yrs followup (range)	9.4 (2.3–18)	3.3 (1.1–7.1)	8.0 (1.1–18)	<0.01 (Wilcoxon rank-sum test)
No. gender (%):				0.68 (Fisher exact test)
Female	39 (42)	14 (47)	53 (43)	
Male	54 (58)	16 (53)	70 (57)	
No. imbrication (%)	24 (26)	8 (27)	32 (26)	>0.99 (Wilcoxon rank-sum test)
No. appendiceal adhesions (%)	37 (40)	14 (47)	51 (42)	0.53 (Wilcoxon rank-sum test)
No. cecal adhesions (%)	21 (23)	10 (33)	31 (25)	0.24 (Wilcoxon rank-sum test)
No. surgical approach (%):				0.83 (Fisher exact test)
Open	40 (43)	12 (40)	52 (42)	
Laparoscopic	53 (57)	18 (60)	71 (58)	
No. procedure (%):				0.78 (Fisher exact test)
ACE	79 (85)	25 (83)	104 (85)	
Urinary diversion	14 (15)	5 (17)	19 (15)	
BMI percentile:				0.49 (Wilcoxon rank-sum test)
Median (range)	87 (1–99)	95 (21–99)	88 (1–99)	
No. pts	87	29	116	
No. BMI category (%):				0.33 (Fisher exact test)
Normal	41 (44)	11 (37)	52 (42)	
Overweight	15 (16)	3 (10)	18 (15)	
Obese	31 (33)	15 (50)	46 (37)	
Unknown	6 (7)	1 (3)	7 (6)	

studies that have evaluated time showed a median time to stenosis of less than 1 year.[5,8,20] It has been postulated that later occurrences may be due to unreported periods of noncompliance. In the present study a minimum of 1 year of followup was required for inclusion. Of cases of stenosis 67% (8 of 12) occurred within 6 months and 75% (9) occurred within 1 year of surgery (Kaplan-Meier curves, fig. 2). Two cases occurred more than 3 years postoperatively. Whether these later occurrences were due to noncompliance could not be determined.

A number of patient and technical variables that may contribute to stenosis have been evaluated in other series, including age, obesity, compliance, stoma location and cecal imbrication. Results have been contradictory. One study suggested increased age at surgery was associated with stenosis,[17] while 2 other studies indicated no association with age.[18,20] In the present study patients with the new stoma technique were on average 2 years older at surgery. Cox proportional hazards analysis of time to stenosis including all measured variables did not

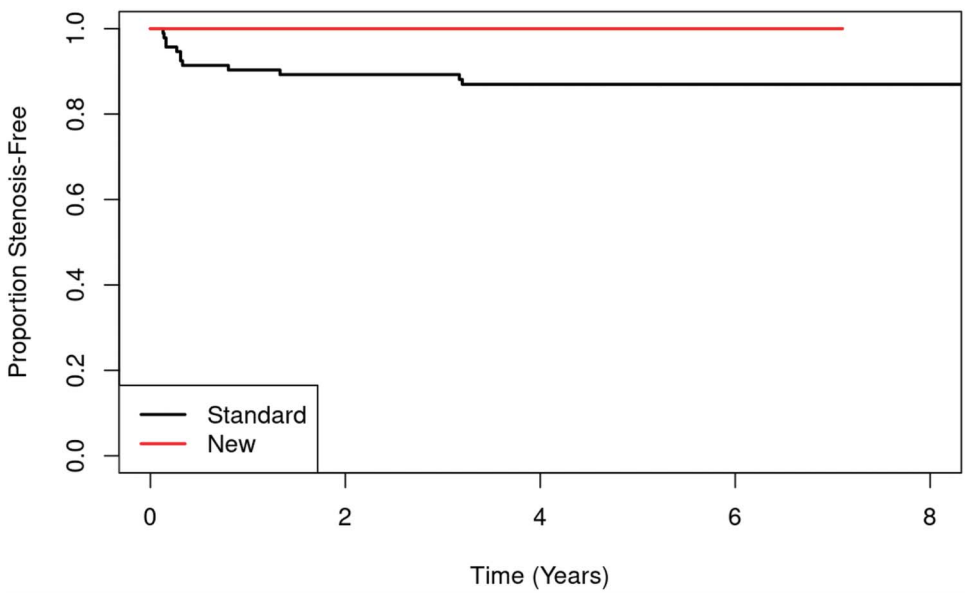

Figure 3. Kaplan-Meier plot of time to stenosis by stoma type

Table 2. *Cox proportional hazards analysis of time to stenosis by patient and surgery characteristics*

	HR (95% CI)	p Value
Gender (male vs female)	0.55 (0.17, 1.64)	0.28
Age at surgery (mos)	0.99 (0.99, 1.01)	0.89
Imbrication (yes vs no)	0.66 (0.13, 2.26)	0.53
Appendiceal adhesions (yes vs no)	0.31 (0.06, 1.08)	0.07
Cecal adhesions (yes vs no)	0.72 (0.14, 2.49)	0.63
Surgery approach (laparoscopic vs open)	1.41 (0.47, 4.91)	0.55
Stoma (new vs standard)	0.13 (0.00, 0.95)	0.04
Urinary diversion vs ACE	1.29 (0.25, 4.46)	0.72
BMI category:		
Overweight/obese vs normal	0.67 (0.21, 2.00)	0.47
Overweight vs normal	0.54 (0.06, 2.47)	0.46
Obese vs normal	0.68 (0.19, 2.14)	0.51

demonstrate age was associated with stenosis ($p=0.89$).

More than 40% of patients with spina bifida are overweight or obese. The appropriate measurement tool for BMI in patients with spina bifida is controversial due to lower limb hypoplasia and vertebral anomalies. Height is suitable for lower level lesions, whereas arm length and other anthropometric measurements improve accuracy for thoracic level.[21,22] In our population, which included nonneuropathic cases, more than 50% of individuals were overweight or obese at surgery. At least 4 institutions have analyzed obesity as a risk factor for stenosis with contradictory results.[9,17,23,24] These studies also included nonappendiceal conduits. Standard BMI calculations with height were used but categorization of obesity differed. In the present study neither obesity nor overweight status was associated with stenosis.

Our preference is to place stomas in the umbilicus since it is a thin exit point from the abdomen and can be hidden. Some patients cannot have an umbilical stoma due to anatomy or placement of 2 stomas. Other investigators have not found location of the stoma to be associated with stenosis.[8,9,17] Cecal plication has been considered critical for stomal continence by some, although Malone suggested it may not be necessary.[16] Our group has studied the association of imbrication with stomal continence and proposed a grading system.[25,26] Despite potential effects on perfusion, this study and another series did not demonstrate an association between cecal imbrication and stenosis.[20]

The group at Indiana University has written extensively on the Malone ACE and urinary diversion. In their large experience they did not find an association between stoma location and stenosis. According to VanderBrink et al, "Potential technical causes contributing to stomal stenosis are excessive tension on the mucocutaneous anastomosis and/or poor blood supply to distal appendix or skin flap."[8] The presented technique might mitigate these 2 factors. The superior holding stitch prevents tension on the anastomosis during the healing phase when the patient moves. Since the distal appendix is not used for the stoma and the blood supply is never violated, perfusion is ensured.

Date of surgery (chronology) was included in the analysis to determine if there was a learning curve. During the 12 years of application of the standard stoma the 12 cases of stenosis were evenly distributed. Statistical analysis did not reveal date of surgery was associated with stenosis.

This study is limited by its retrospective nature and lack of randomization. A single surgeon experience carries inherent biases of technique and could constitute an advantage or disadvantage when evaluating an isolated technical change. Although every known variable that could impact stenosis was evaluated, there are certainly factors that are unknown.

CONCLUSIONS

Similar to other studies, this analysis did not show an association of patient age, BMI, gender or cecal imbrication with stenosis. Other variables that have not been previously evaluated, such as appendiceal position, adhesions and approach, did not impact the incidence of stenosis. The only factor that was observed to be associated with stenosis was procedural completion with the standard vs new technique. Stomal stenosis of appendicostomy may be lessened by preservation of the distal appendiceal vasculature and tip, and opening the lumen more proximally.

ACKNOWLEDGMENT

Dr. Blythe Durbin-Johnson performed statistical analyses.

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EDITORIAL COMMENTS

Stomal stenosis following appendicovesicostomy or the Malone ACE procedure has been the scourge of many a reconstructive surgeon. Inadequate blood supply is often cited as the primary reason for this unfortunate outcome. Efforts to avoid tissue compromise include maximal preservation of vascularity and performing a tension-free spatulated anastomosis to a wide based skin flap. The author describes an ingenious method for preserving blood supply to the conduit and, in the series presented, demonstrates improvement in conduit functionality.

While preservation of blood supply is perhaps the most important variable in determining the incidence and prevalence of stomal stenosis, other factors may have an important role. Minimizing stretch on the conduit blood supply by anchoring the bladder or cecum to the rectus, documentation of initial conduit suitability (ie length and diameter) and a standardized, unbiased definition of stomal stenosis are a few additional variables that should be considered to more fully validate the benefit of this novel technique.

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Quality improvement is a frequently used term in health care organizations. Improving patient outcomes is what all of us aspire to while practicing medicine. Deming proposed a 4-step method for continuous improvement of a process called PDSA (Plan-Do-Study-Act).[1] In the model a process is observed and a course of action is determined to improve it (Plan), the plan is enacted (Do), the results are analyzed (Study) and the results influence the process (Act). Implementing such methodology can change clinical outcomes, and this blueprint appears to have been followed in the current study.

Stomal stenosis following creation of continent catheterizable channels is frequently encountered within 2 years (reference 7 in article). The author examined the rate of stomal stenosis with 1 technique of Malone ACE procedure. Here he reports a new technique predicated on preservation of the

appendiceal tip and vessels while opening the lumen in a more proximal and vascular area, aimed at minimizing stomal stenosis. This innovative procedure is based on sound anatomical principles respecting the delicate vasculature of the appendix. The author used this technique and analyzed the surgical outcomes in 30 patients after a median followup of 3 years. I applaud the author for presenting the outcomes after adequate time elapsed, when one would feel confident in not "speaking too soon" to report on stomal stenosis. The results are striking in that stenosis was eliminated with the new technique. This outcome could be characterized as the ultimate in quality improvement and this technique should be considered by surgeons who perform the Malone ACE procedure.

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