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ORIGINAL ARTICLE

Fact or myth? The long shared common wall between the fistula and the urethra in male anorectal malformation with urethral bulbar fistula

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Abstract

Aim It has long been considered surgical dogma that the length of the shared common wall (CW) between a fistula and the urethra in males with anorectal malformation (ARM) and rectourethral bulbar fistula (RUBF) is considerably longer than in males with ARM and rectourethral prostatic fistula (RUPF). This belief has led surgeons who perform laparoscopic-assisted anorectoplasty (LAARP) for RUPF to avoid LAARP for RUBF for risk of potential injury to the urethra or incomplete removal of the fistula. In this study, we compared CW between RUBF and RUPF using distal colostography (DCG) and direct intraoperative measurements.

Methods DCG of rectourethral fistula patients ($n=63$; RUBF: $n=44$; RUPF: $n=19$) were used to measure CW retrospectively. Results were expressed as a ratio of the height of L4; i.e., CW:L4. If less than 0.7, the CW was classified as being "short"; if 0.71–1.4, as being "medium"; and if greater than 1.41, as being "long". CW that could not be measured was classified as indeterminate. 24 of these patients also had CW measured intraoperatively during LAARP as previously described. The results obtained using both techniques were also compared.

Results Surprisingly, CW:L4 in RUBF patients was short in 47.7%, medium in 27.3%, long in 20.5%, and indeterminate in 4.5% on DCG, equivalent to mean lengths of 7 mm, 8.5 mm, and 10.3 mm obtained using direct intraoperative measurement for short, medium, and long CW:L4 categories, respectively. CW:L4 in RUPF was short in 73.6%, medium in 10.5%, and long in 5.2% on DCG, while mean intraoperative measurements were 5 mm, 7 mm, and 10 mm, respectively. Differences in CW measured intraoperatively were not significantly different between RUBF and RUPF ($p=$NS).

Conclusion From our findings, 47.7% of CWs in RUBF were short using two independent methods, with only 20.5% being long. Thus, LAARP should be considered actively for treating selected RUBF cases and not be excluded on the basis of CW length.

Keywords Length of common wall · Rectourethral bulbar fistula · Rectourethral prostatic fistula · Anorectal malformation

Introduction

Since laparoscopically assisted anorectoplasty (LAARP) was described by Georgeson et al. [1] as an alternative to posterior sagittal anorectoplasty (PSARP) for the surgical treatment of rectourethral fistula in male patients with high-/intermediate-type imperforate anus, many pediatric surgeons have considered it to be too technically challenging to perform despite growing interest in the procedure.

It has long been considered surgical dogma that the length of the shared common wall (CW) between the fistula and urethra in males with anorectal malformation (ARM) and rectourethral bulbar fistula (RUBF) was considerably longer

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than in males with ARM and rectourethral prostatic fistula (RUPF). This belief has influenced surgeons who perform LAARP for RUPF to avoid LAARP for RUBF because of the purported greater risk for potential injury to the urethra and risk for incomplete removal of the fistula.

In this study, we compared CW between RUBF and RUPF cases using distal colostography (DCG) and direct intraoperative measurement.

Materials and methods

The medical records of 63 male ARM with rectourethral fistula cases treated at two institutions (Juntendo University Hospital and Cedars-Sinai Medical Center) were reviewed retrospectively. All patients were diagnosed as neonates and had colostomies, and distal colostography (DCG) and voiding cystourethrography (VCUG) to evaluate anorectal anatomy. Of the 63 patients, 39 had posterior sagittal anorectoplasty (PSARP) and 24 had laparoscopic-assisted anorectoplasty (LAARP). PSARP were performed according to the technique described by de Vries and Pena [2] and all LAARPs were performed according to the conventional technique described by Georgeson et al. [1]. All surgery was performed under the direct supervision of one of two board-qualified pediatric surgeons with extensive experience of both techniques (PF or AY).

Measuring CW using DCG

CW was measured retrospectively using DCG and expressed as a ratio of the height of L4 vertebral body, CW:L4. If less than 0.7, CW was classified as being "short"; if 0.71–1.4, CW was "medium"; and if greater than 1.41, as "long" (Fig. 1). CWs that could not be measured were classified as indeterminate.

Measuring CW during LAARP

CW was measured directly during LAARP using a modification of a technique that has been published elsewhere previously [3]. Briefly, the fistula is dissected carefully close to the urethra and laid open. A fine catheter with 10 mm calibration is inserted by the laparoscopic surgeon until it

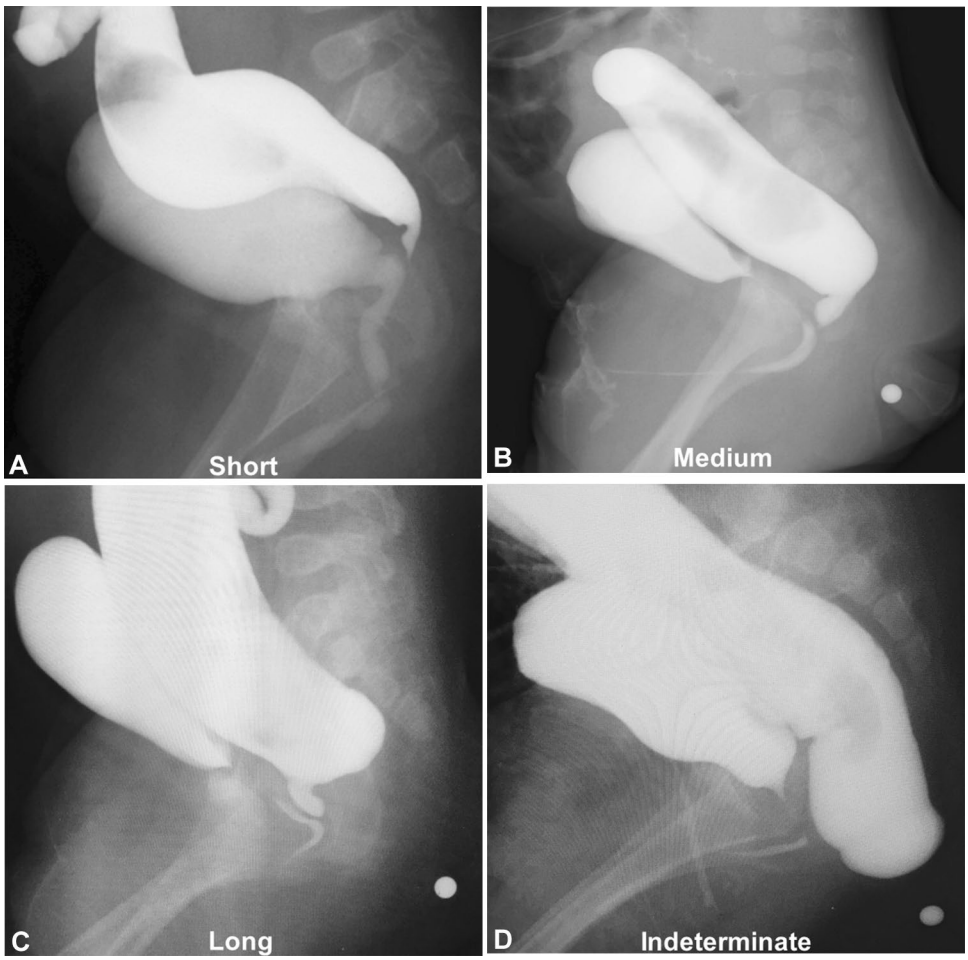

Fig. 1 Common wall lengths (CW) measured on distal colostography expressed as a ratio of the height of L4. **a** < 0.7 = "short", **b** 0.71–1.4 = "medium", **c** > 1.41 = "long", **d** "indeterminate"

is seen to emerge into the urethra by another surgeon performing cystoscopy. The laparoscopic surgeon measures the distance from the point where dissection was ceased at the rectal end to the urethral orifice. The fistula is then separated from the urethra creating a submucosal plane and dissected further distally for the distance just measured. Residual rectourethral fistula measurement can be repeated if necessary with further dissections until the fistula is less than 5 mm in length. Once less than 5 mm in length, the fistula is excised and ligated with an Endoloop (Ethicon, OH). We used the distance to the submucosal plane as the direct measurement of CW length (Fig. 2).

The Student's *t* test was used for statistical analysis. A *p* value of less than 0.05 ($p < 0.05$) was considered statistically significant.

This study was approved by the Juntendo University School of Medicine Institutional Review Board and complies with the Helsinki Declaration of 1975 (as revised in 1983).

Results

DCG and VCUG clearly defined the location of fistulas in all cases. They were RUBF ($n = 44$) and RUPF ($n = 19$). CWs measured on DCG are summarized in Table 1. CW:L4 was short in 21/44 RUBF cases and 14/19 RUPF cases (47.7% versus 73.7%); medium in 12/44 RUBF and 2/19 RUPF cases (27.3% versus 10.5%); long in 9/44 RUBF cases and 1/19 RUPF cases (20.5% versus 5.3%), and indeterminate in 2/44 in RUBF and 2/19 RUPF cases (4.5% versus 10.5%).

Table 1 Number of cases in each length category according to fistula type ($n = 63$)

CW:L4 ratio[a]	RUBF	RUPF
Short (< 0.7)	21	14
Medium (0.71–1.4)	12	2
Long (> 1.41)	9	1
Indeterminate	2	2

CW common wall, *RUBF* recto-urethral bulbar fistula, *RUPF* recto-urethral prostatic fistula

[a]Definition: length of the common wall as a ratio of the height of L4 measured using distal colostography

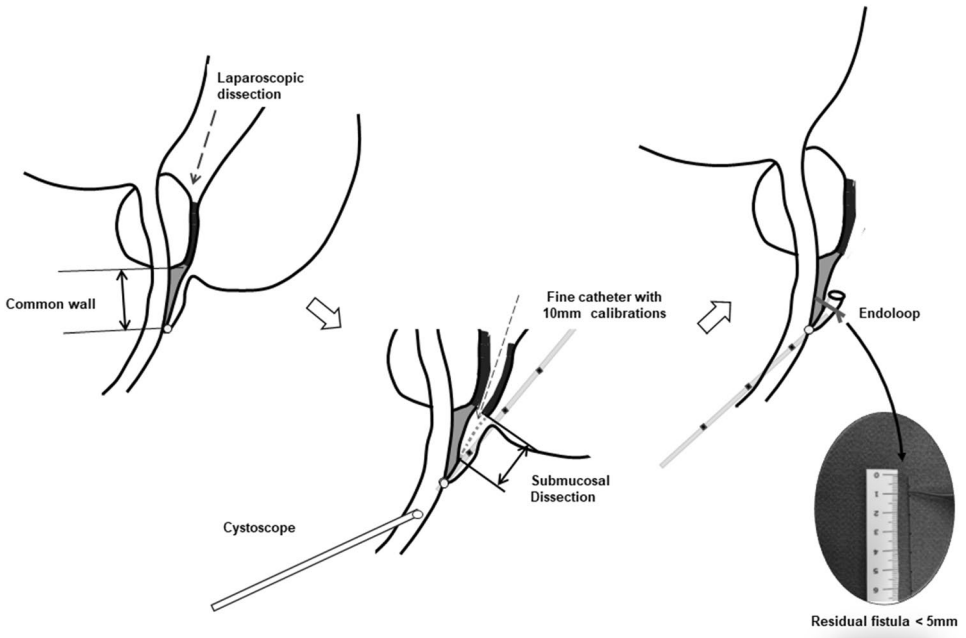

Fig. 2 The fistula is dissected carefully close to the urethra and laid open. A fine catheter with 10 mm calibrations is inserted by the laparoscopic surgeon until it is seen to emerge into the urethra by another surgeon performing cystoscopy. The laparoscopic surgeon measures the distance from the point where the dissection ends at the rectal end to the urethral orifice. The fistula is then separated from the urethra creating a submucosal plane and dissection proceeds further distally for the distance just measured. Residual rectourethral fistula measurement can be repeated if necessary with further dissection until less than 5 mm. Once less than 5 mm in length, the fistula is excised and ligated. Then, the actual length of the common wall is calculated as the distance to the submucosal plane. We used the distance to the submucosal plane as the direct measurement of CW length

Intraoperative CW measurements are shown in Table 2. Of the total of 24 LAARP cases who had intraoperative measurement, mean length in short CW:L4 cases was 7.0 mm (range 5–12 mm) in RUBF ($n=8$) and 5.0 mm (range 4–6 mm) in RUPF ($n=5$); in medium CW:L4 cases was 8.5 mm (range 8–10 mm) in RUBF ($n=4$) and 10.0 mm in RUPF ($n=1$); in long CW:L4 cases was 10.3 mm (range 8–15 mm) in RUBF ($n=4$) and 10.0 mm in RUPF ($n=1$); and in the only indeterminate CW:L4 case measured intraoperatively was 10.0 mm in RUPF. The overall mean CW measured intraoperatively was 8.1 (range 5–15 mm) in RUBF ($n=16$) and 6.5 (range 4–10 mm) in RUPF ($n=8$); p = NS.

Interestingly, for RUBF, 47.7% were short, 27.3% were medium, 20.5% were long and 4.5% were indeterminate on DCG. Intraoperative measurements followed the same trend with short CW:L4 having a mean length of 7 mm, medium CW:L4 having a mean length of 8.5 mm, and long CW:L4 having a mean length of 10.3 mm. For RUPF, 73.7% were short, 10.5% were medium, and 5.0% were long on DCG. Intraoperative measurements again followed the same trend with short CW:L4 having a mean length of 5.0 mm, medium CW:L4 having a mean length of 7.0 mm, and long CW:L4 having a mean length of 10.0 mm. Differences in CW measured intraoperatively between RUBF and RUPF were not significant ($p=$ NS).

Discussion

Laparoscopy to repair high/intermediate ARM in male infants has been widely used by pediatric surgeons, in an effort to make surgery less invasive and more cosmetic, as well as enhance recovery with less postoperative pain and enable patients to achieve reliable bowel function. However, LAARP is still not recommended for RUBF in the literature [4, 5], because of a seemingly higher risk for posterior urethral diverticula. In our experience, outcomes after LAARP are comparable to PSARP [6–10]. The procedure has its challenges regarding dissection and ligature of the fistula, because the urethra and rectum share a common wall, which could pose a significant risk for injuring the urethra. In fact, as the surgeon proceeds with dissection of the distal rectum, separating the anterior rectal wall from the posterior urethral wall will become more difficult. In addition, the lowest part of the rectum will be particularly challenging to dissect; consequently, the surgeon may be tempted to transect the rectum, leaving the distal portion attached to the bulbar urethra. On the other hand, some surgeons may opt dissecting closer to the rectum to avoid an intraoperative urethral injury, which may increase the risk for incomplete excision of CW, contributing to subsequent diverticulum formation [11, 12]. In fact, most pediatric surgeons would concede that it is safer to risk a potential diverticulum due to incomplete excision than injure the urethra and cause stenosis by dividing CW too close to the urethra.

In the literature, there are only a few reports about modified fistula dissection, involving dissection of the distal fistula along the submucosal plane up to where the fistula joins with the urinary tract, performed during LAARP to prevent possible diverticulum formation [5, 13]. Rollins et al. described simple division of a fistula and placement of an indwelling urethral catheter without using clips or sutures to lower risks for injuring the urethra. It is easy to imagine worst case scenarios for these techniques with dissection progressing by trial and error at best and blindly at worst, because the surgeon essentially has no idea of the distance between the rectal and urethral orifices of a fistula. Thus, by measuring the length of a fistula with a cystoscope and a calibrated catheter during LAARP as previously reported [3], the surgeon can dissect a fistula distally with confidence and without any risk for injury to genitourinary structures. In support of this approach, we use magnetic resonance imaging to investigate for posterior urethral diverticulum formation during routine follow-up, and to date have had no incidence, even in RUBF cases [14].

In this series, we found CW:L4 to be short in 47.7% of RUBF cases with a mean length of 7.0 mm (range 5–12 mm), and long in only 20.5% of RUBF cases with a mean length of 10.3 mm (range 8–15 mm), which casts doubts on the established concept that CW is longer in RUBF. Mean intraoperative CW measured during LAARP in RUBF ($n=16$) and

Table 2 Common wall lengths measured directly during LAARP ($n=24$) according to CW:L4 categories

CW:L4 category (RUBF/RUPF)	Mean RUBF length (range)[a]	Mean RUPF length (range)[a]
Short: ($n=8/5$)	7.0 (5–12)	5.0 (4–6)
Medium: ($n=4/1$)	8.5 (8–10)	7.0
Long: ($n=4/1$)	10.3 (8–15)	10.0
Indeterminate: ($n=0/1$)	NA	10.0
	Overall: 8.1 (5–15)	Overall: 6.5 (4–10)

LAARP laparoscopic-assisted anorectoplasty, *RUBF* recto-urethral bulbar fistula, *RUPF* recto-urethral prostatic fistula; *NA* not available

[a]mm measured directly during LAARP

RUPF ($n=8$) also followed the same pattern. In fact, Pena et al. described that the rectum is separated from the urethra by a submucosal plane of separation that is approximately 5–10 mm above a fistula, which would be equivalent to CW [15] that supports our findings. Thus, the length of CW is more likely to be of the same order in both RUBF and RUPF, rather than being longer in RUBF compared with RUPF. What this means is that the long CW in RUBF is actually a myth. The long CW has been an issue for treating RUBF, but its location deep in the pelvis nevertheless still makes surgical intervention for RUBF extremely challenging.

To the best of our knowledge, this is the first report about measuring CW in male patients with high/intermediate type ARM and RUBF or RUPF according to two different techniques, DCG and direct measurement and comparing them independently. Our findings would suggest that LAARP could be used for the safe excision of all rectourethral fistulas, even RUBF, in contrast to other reports in the literature. In fact, with accurate measurement and controlled dissection, the only conceivable risk for injury would be an accident or something completely unexpected. Although the number of cases in this series is limited, our findings would suggest that LAARP should not be excluded as a procedure for treating selected ARM patients with RUBF on the basis of CW length. A larger series is planned to provide further evidence to support our findings.

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