TITLE PAGE

Title:

Surgical management of rectoprostatic and rectobulbar anorectal malformations

Running head:

Laparoscopic- assisted and posterior sagittal anorectoplasties compared

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ABSTRACT

Purpose: Laparoscopically assisted anorectoplasty (LAARP) was compared to posterior sagittal anorectoplasty (PSARP) in treatment of male imperforate anus associated with either recto-prostatic fistula (RPF) or recto-bulbar fistula (RBF).

Method: 19 RPF patients (12 treated by LAARP and 7 by PSARP) and 26 RBF patients (14 treated by LAARP and 12 by PSARP) between 1995 to 20014 were retrospectively assessed using a fecal continence evaluation questionnaire (FCE) (with a maximum score of 10), a FCE score coefficient variation, as well patients' MRI scores, anorectal angle values (AA), and incidence of postoperative complications. Statistical significance was determined at $p<.05$.

Results: Both groups were similar in mean age and mean weight at repair, as well as sacral status. Postoperatively, mean MRI scores, mean AA, and biochemistry were also similar (p=NS). All cases treated with LAARP showed consistently higher and less variable FCES values, fewer wound infection incidence, but greater rectal mucosal prolapse unrelated with sacrum status. Significantly lower doses of postoperative analgesia were needed in all LAARP cases ($p< .05$).

Conclusion: Technical outcomes appear to be similar based on imaging studies, but FCES assessed functional outcomes appear to favor LAARP for treatment of both RPF and RBF.

Keywords: laparoscopy-assisted anorectoplasty, posterior sagittal anorectoplasty, imperforate anus, recto-bulbar fistula, recto-prostatic fistula

INTRODUCTION

Since laparoscopically assisted anorectoplasty (LAARP) was described by Georgeson et al as an excellent alternative to posterior sagittal anorectoplasty (PSARP) [1] , as of late LAARP has become an increasingly common surgical treatment of choice for male patients with high-/intermediate-type imperforate anus (IA). Several reports comparing LAARP and PSARP have favored LAARP, describing excellent visibility of the rectal fistula, accurate placement of the pull-through segment at the center of the pelvic muscle complex without the need for dividing muscles, and satisfactory fecal continence as advantages over PSARP [2-4]. However, above mentioned reports include a variety of IA cases with recto-vesical, recto-prostatic, recto-bulbar, recto-vaginal, or absent fistulas, or cloaca, making any conclusions about comparison of surgical techniques difficult to apply because of the wide spectrum of bowel continence issues associated with mentioned variety of IA. Expectations that outcomes after LAARP may be better than outcomes after PSARP are in general, but direct comparison is hindered by subject anatomical i.e. clinical differences, resulting in few reports getting into details about any LAARP advantages over PSARP.

We focused on only recto-prostatic fistula (RPF) or recto-bulbar fistula (RBF) to best compare LAARP with PSARP.

MATERIALS and METHODS

We reviewed 45 male high-type imperforate anus (MHIA) cases (19 RPF and 26 RBF) treated from 1995 to 2015 at our institution, prospectively. All patients had been treated initially

as neonates and had colostomies done. Of the 19 RPF patients, 12 were treated with LAARP and 7 were treated with PSARP. Of the 26 RBF patients, 14 were treated with LAARP and 12 were treated with PSARP. All repairs were performed under the direct supervision of a single board-qualified specialist pediatric surgeon (AY) with extensive experience in both techniques and success rates equivalent to published reports [5-8]. All LAARP were performed in accordance with the conventional technique as described by Georgeson et al [1] and all PSARP were performed in accordance to the technique as described by de Vries and Pena [9] . In this series, six PSARP surgeries were performed on RBF patients before LAARP was introduced and six PSARP surgeries were performed after LAARP was introduced first. We continued performing PSARP after LAARP was introduced in six cases where complications secondary to residual fistula during LAARP were experience [10]. However, this lead to our refining of the LAARP procedures done in that we developed a novel technique to measure the length of the distal fistula before excising it [11] ; that improved our confidence with regard to LAARP used. Postoperative analgesia was administered according to our routine postoperative pain protocol [12] ; all cases were commenced initially on a fentanyl infusion at 0.5-1μg/kg/hr.

Measuring the anorectal angle (AA)

The anorectal angle is the angle formed between the rectum and the anal canal during barium enema, and results are expressed in degrees [6]. The axis of the anal canal was determined by drawing a line down the middle of the anal canal and the axis of the rectum by drawing a line parallel to the most inferior portion of the rectum just cephalad to the anal canal. Barium enema was administered with the patient at rest, in the lateral decubitus position for patients from 6 months to 4 years in age after they had undergone anorectoplasty; this was dome

for all 45 subjects.

Pelvic magnetic resonance imaging score (MRI score)

Magnetic resonance imaging (MRI) was performed on patients of ages between 6 months and 10 years old postoperatively in all of the 45 cases in this series to evaluate each patient's anorectal anatomy, as has been done previously [5]. MRI images were obtained in sagittal and axial planes (parallel to pubococygeal line) as contiguous 4 to 7 mm slices along the anal canal and rectum. MRI images were assessed by a reviewer of consultant level who was blinded to the type of operation done. The muscle thickness at the levels of the external sphincter (ES) and puborectalis (PR)were noted for symmetry and graded as good, fair or poor depending on whether muscle mass was obvious, thin, or virtually invisible. In order to qualify for poor, the muscles had to be either absent or virtually invisible.

Scoring was done as follows: if muscles appear to be symmetrical and their thickness was good-good at both 3 and 9 0'clock positions, the score was 0. Slight difference in thickness (i.e., good-fair or fair-poor) scored 1 point, and marked difference (i.e., good-poor) scored 2 points. Hence the worst score for each of the ES and PR levels was 2 to give the worst total score of 4 (i e ES = 2 +PR = 2).

Fecal continence evaluation questionnaire score (FCE score)

We developed a structured fecal continence evaluation (FCE) questionnaire [5, 6] that we administer routinely at follow-up outpatient visits to assess bowel function in all of our patients who have undergone surgery that might affect defecation. Briefly, 5 parameters (frequency of defecation, incidence of staining/soiling, presence of perianal erosion, anal shape, and

requirement for medication) are measured on a scale of 0 to 2 per parameter, for a total highest score value of 10. In the questionnaire, we use staining to refer to fecal stains on undergarments and soiling to mean actual feces on undergarments. In the present series, we excluded cases followed up for less than 3 years, cases where mental retardation is present, and cases with chromosomal anomalies or sacral anomalies such as tethered cord. Thus, FCE data were obtained for 36 cases (16 RPF: 9 LAARP and 7 PSARP; 20 RBF: 10 LAARP and 10 PSARP) and scored by a single surgeon (AY) resulting in FCE scores. Mean annual FCE score values were determined and compared between the two groups. The coefficient of variation (CV; ratio of standard deviation to mean) was calculated for evaluating the degree to which FCE score are at variance (FCESCV).

Surgical stress

To assess surgical stress, mean peak white cell counts (WCC; normal<10,000 $\times 10^6$/L), the difference in duration of elevated WCC, peak serum C-reactive protein values (CRP; normal<0.3mg/dL), and the difference in duration of elevated CRP levels between the two groups were compared.

Data gathered included age and weight at repair, MRI score values, AA, incidence of complications, indicators of surgical stress, dosages of postoperative analgesia needed, as well as FCE score and FCESCV values. Data were expressed as means ± standard deviation values. The Student's *t* test and Chi-squared test values were used for statistical analysis. A *p* value of less than 0 .05 ($p<.05$) 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 (revised 1983).

RESULTS

Mean ages at repair were similar according to type of fistula (RPF: LAARP vs. PSARP 7.6±3.0 months vs. 4.0±3.5 months; RBF: LAARP vs. PSARP 8.1±4.0 months vs. 8.2±5.1 months, respectively. *p*=NS). Mean lengths of follow-up after repair were significantly longer for both RPF and RBF patients treated with PSARP compared to those treated using LAARP (RPF: LAARP vs. PSARP 99.3±55.5 months vs. 261.0±49.2 months; RBF: LAARP vs. PSARP 76.6±56.9 months vs. 148.0±56.1 months, respectively. $p<.001$). Mean operative times for both RPF and RBF patients treated with LAARP were significantly longer than when PSARP was used (RPF: LAARP vs. PSARP 494±113 minutes vs. 335.8±92.3 minutes; RBF: LAARP vs. PSARP 403±80 minutes vs. 206±31 minutes, respectively. $p<.001$). The sacral ratio was <0.4 (LAARP vs. PSARP: RPF n =0 vs. n=1; RBF n =2 vs. n=0), 0.4-0.7 (RPF n=6 vs. n=5; RBF n=9 vs. n=7), and >0.7 (RPF n=6 vs. n=1; RBF n=3 vs. n=5). The status of the spinal cord was normal (LAARP vs. PSARP: RPF n =12 vs. n=6; RBF n =12 vs. n=11) and tethered (RPF n=0 vs. n=1; RBF n=2 vs. n=1). Mean MRI scores were not significantly different (RPF: LAARP vs. PSARP 0.7±0.7 vs.1.3±0.8; RBF: LAARP vs. PSARP 0.9±0.5 vs. 0.7±0.7, respectively. *p*=NS), indicating that there were no differences in muscle thickness when one or the other repair technique was applied.

Mean annual FCE scores for the first seven years after surgery (at patients' ages 3 to 9) for each group were: 7.3±0.8 (n=9), 7.6±0.8 (n=8), 8.0±0.9 (n=5), 8.3±0.7 (n=5), 8.5±0.7 (n=4) ,

Figure1

8.8±0.5 (n=4), and 8.8±0.3 (n=4) for LAARP-RPF; 6.4±1.5 (n=7), 6.6±1.3 (n=7), 7.3±1.1 (n=7), 7.4±1.1(n=7), 7.4±0.9 (n=7), 7.4±0.9 (n=7), and 7.4±0.7 (n=7) for PSARP-RPF (Figure 1); 7.8±0.4 (n=10), 7.9±0.6 (n=8), 8.4±0.5 (n=7), 8.8±0.3 (n=4), 8.9±0.3 (n=4), 9.0±0.4 (n=4) and 9.1±0.3 (n=4) for LAARP-RBF; and 7.6±1.5 (n=10), 7.8±1.5 (n=10), 8.2±1.4 (n=10), 8.3±1.6 (n=10), 8.5±1.3 (n=9), 8.6±1.1 (n=7), and 8.8±1.3 (n=5) for PSARP-RBF (Figure 2). While LAARP resulted in consistently higher scores in both RPF and RBF patients for the duration of the study, FCE scores were only statistically significant five, six and seven years after repair in RPF patients, while they were not statistically significant at any time in RBF patients.

Figure2

FCESCV values were found to be smaller in LAARP treated patients compared to those in PSSARP treated patients for both RPF and RBF cases (LAARP vs. PSARP. RPF: 0.10 vs. 0.23, 0.10 vs. 0.20, 0.11 vs 0.15, 0.08 vs 0.15, 0.08 vs 0.12, 0.05 vs 0.12 and 0.03 vs 0.09; RBF: 0.05 vs 0.20, 0.07 vs 0.19, 0.06 vs 0.17, 0.03 vs 0.19, 0.03 vs 0.15, 0.04 vs 0.12 and 0.03 vs 0.14 respectively) (Figure 3).

Figure3

Mean peak WCC and duration of elevated WCC for LAARP-RPF patients and PSARP-RPF patients were 15,100±4,010 $\times 10^6$/L and 3.2±0.6 days versus 12,000±2,603 $\times 10^6$/L and 3.0±1.1 days, respectively (all *p*=NS) and for LAARP-RBF and PSARP-RBF were 13,260±4,015 $\times 10^6$/L and 3.0±1.6 days versus 12,900±2,100 $\times 10^6$/L and 4.2±4.0 days, respectively (all *p*=NS). Mean peak CRP levels and duration of elevated CRP levels for LAARP-RPF and PSARP-RPF patients were 8.7±3.8mg/dL and 5.4±0.9 days versus 5.7±1.0mg/dL and 4.3±1.0 days, respectively (all *p*=NS) and for LAARP-RBF and PSARP-RBF were 5.9±2.1mg/dL and 4.5±2.6 days versus 6.2±2.2mg/dL and 5.4 ± 4.8 days, respectively (all *p*=NS). Mean duration of needed postoperative analgesia in all LAARP patients was significantly lower than in the PSARP

patients; LAARP-RPF vs. PSARP-RPF: 1.7±0.7 days vs. 4.2±0.9 days; LAARP-RBF vs. PSARP-RBF: 1.6±0.6 days vs. 3.8±1.2 days, respectively. $p<.05$).

As for complications, there were nine cases of postoperative rectal mucosal prolapse, and one case of enlarged residual fistula as previously reported [10] in LAARP treated patients; two cases of postoperative rectal mucosal prolapse, three cases of wound infection incidence, and one case of anal stenosis in PSARP treated patients. There was no incidence of infection in LAARP treated patients.

DISCUSSION

This is the first official report comparing LAARP and PSARP performed under the supervision of the same and single surgeon in a series of MHIA patients with RPF and RBF matched for demographics and incidence of sacral anomalies. By minimizing as many of IA variables as possible, we believe that we have been able to clearly compare techniques and outcomes, more directly than ever previously reported in the literature; thus the present study is the first of its kind in which the outcomes of LAARP or PSARP repair in MHIA patients with RPF or RBF have been compared to date.

It has been known that postoperative fecal continence is dependent on the type of malformations present as well as the neurological status of the sacrum and factors specific to very young children. Sacral dysplasia and abnormal sphincters were related to poor prognosis following repair [13, 14]. There have interestingly been no severe sacral abnormalities such as hemivertebrae and vertebral fusions in the present series. And it is also known that the surgical method of anorectal reconstruction may be a significant prognostic factor. Our group has

previously reported IA-LAARP repair to have consistently better outcomes, using a FCE score questionnaire and surgical stress marker readings, when compared to PSARP [5] , although the previous study was narrower in scope than the present one and included variety of IA. The present series as described herein had longer follow-up times compared to the previous one, and included matched cases. FCE score values after LAARP repair were consistently higher although there was no significant difference pertaining to repair technique used in RPF and RBF patients, suggesting that LAARP repair should result in better midterm postoperative fecal continence outcomes, more specifically, the regularity of motions, and lesser staining/soiling; the outcomes were statistically significant seven years after surgery in RPF patients, especially when compared to LAARP repair used for staining /soiling issues. Wong et al [15]compared fecal continence according to surgical procedure and reported that significantly more patients treated with LAARP showed acceptable defecation status; however, mentioned study subjects were evaluated only for frequency of motions as an indicator of fecal continence, which we consider to be insufficient compared to our comprehensive FCE score method. Some reports found that both LAARP and PSARP treated patients had similar bowel movement habits [3, 4]. However, in such studies, the ages of patients at the time of evaluation for fecal continence were significantly lower in the LAARP treated group than the ages of patients treated with PSARP. As functional results improve with time, LAARP appears to deliver better outcomes. Our results demonstrate that FCE score values in RBF and RPF patients following LAARP repair were found to consistently improve overtime, as follow-up also supported this findings.

We found LAARP to be equally as effective as PSARP in both RPF and RPF patients as reported previously [5, 6]. In addition, MRI scores confirmed that pelvic floor muscles in both RBF and RPF patients were found to surround the pull-though canal evenly regardless of

surgical procedure used. In other words, the pull-though canal was perfectly centered in the pelvic floor muscles irrespective of procedure used. However, MRI alone was not reliable for measuring muscle thickness, since used only for semiquantitative assessment because subjective factors would appear to influence the interpretation of MRI images. At the present time, determining muscle thickness quality still presents a challenge since it is difficult to reconstruct fine 3D images of the pelvic muscle complex using the existing MRI technology. Further technological improvements are necessary.

Our analysis shows that LAARP has lower FCESCV than PSARP, a reflection of the precision with which the pulled-through segment can be centered in the pelvic muscle complex without dividing any muscles. This reliability is directly related to preservation of the pelvic floor and is almost expected from LAARP as a procedure. Pena et al reported that complete division of sphincter mechanisms followed by meticulous reconstruction has not been detrimental in their experience [16], however, because PSARP requires division of pelvic floor and sphincter muscles such as the puborectalis, there could be some functional disruption of the contraction function that could contribute to fecal incontinence. Iwanaka et al [17] recommended LAARP as an alternative procedure to PSARR, since the latter was known to cause damage to the sphincter muscles as well as the tiny nerves that maintain anorectal sensation and motility, due to the large incision it requires at the pelvis. Yang et al [18] reported that LAARP significantly improved the anal canal resting pressure and shortened the patient's hospital stay, since LAARP reflected the effect of avoiding injuring the muscle complex and the nerves around the puborectal muscles; thus LAARP preserved better sphincteric function. Such findings match our findings. Our group also believes that a true determinant of outcomes is the quality of the patient's nerves and muscles in the pelvis. LAARP would appear to not damage

nerves and muscles in the pelvis. Fecal continence in both RBF and RPF patients following LAARP repair was found to be less dispersive and stable when compared to continence in both types of patients who had undergone PSARP repair. PSARP appears to be a difficult technique to learn how to use and with less stable outcomes.

Rectal prolapse has been reported in 9-46% of patients following LAARP repair [3, 4]. Other studies have shown that rectal prolapse occurs following PSARP repair at an incidence of 3%. It is more common in patients with higher incidence of malformations and with poor sacral and pelvic musculature, but the higher incidence of postoperative rectal mucosal prolapse after LAARP repair may be secondary to division of anatomic attachments between the posterior rectum and the sacrum during dissection of the rectum. Rectal prolapse was found in 37% of the patients (9/24) in our series after LAARP repair. Prolapse occurrences in our patients were observed only in some of the early cases and may have arisen due to too extensive or aggressive dissection that may have caused disruption of blood perfusion and poor tissue healing, or because the rectum may have been inadequately fixed. The incidence of rectal mucosal prolapse has decreased because of meticulous dissection used and rectal fixation with seromuscular sutures between the rectum and the presacral fascia.

Comparisons were made between the duration in times of elevated levels of white blood cell counts, and peak C-reactive protein levels as indicators of surgical stress in both procedures, and no significant difference was found between LAARP and PSARP. Wound infection incidence was less common in patients where LAARP was used, although operative times in LAARP patients was significantly longer since the procedure is performed through only small 3-5 mm incisions in order to minimize pain, postoperative analgesia dosages were also lower.

In conclusion, to the best our knowledge, this is the first report comparing LAARP and PSARP as treatments in MHIA patients with RPF or RBF. LAARP appears to be associated with less physical impact and less FCESCV implying that LAARP may be more reliable for repairing MHIA with RPF or RBF, despite the risk of higher incidence of anal mucosal prolapse.

Disclosures

The authors have no conflicts of interest or financial ties to disclose.

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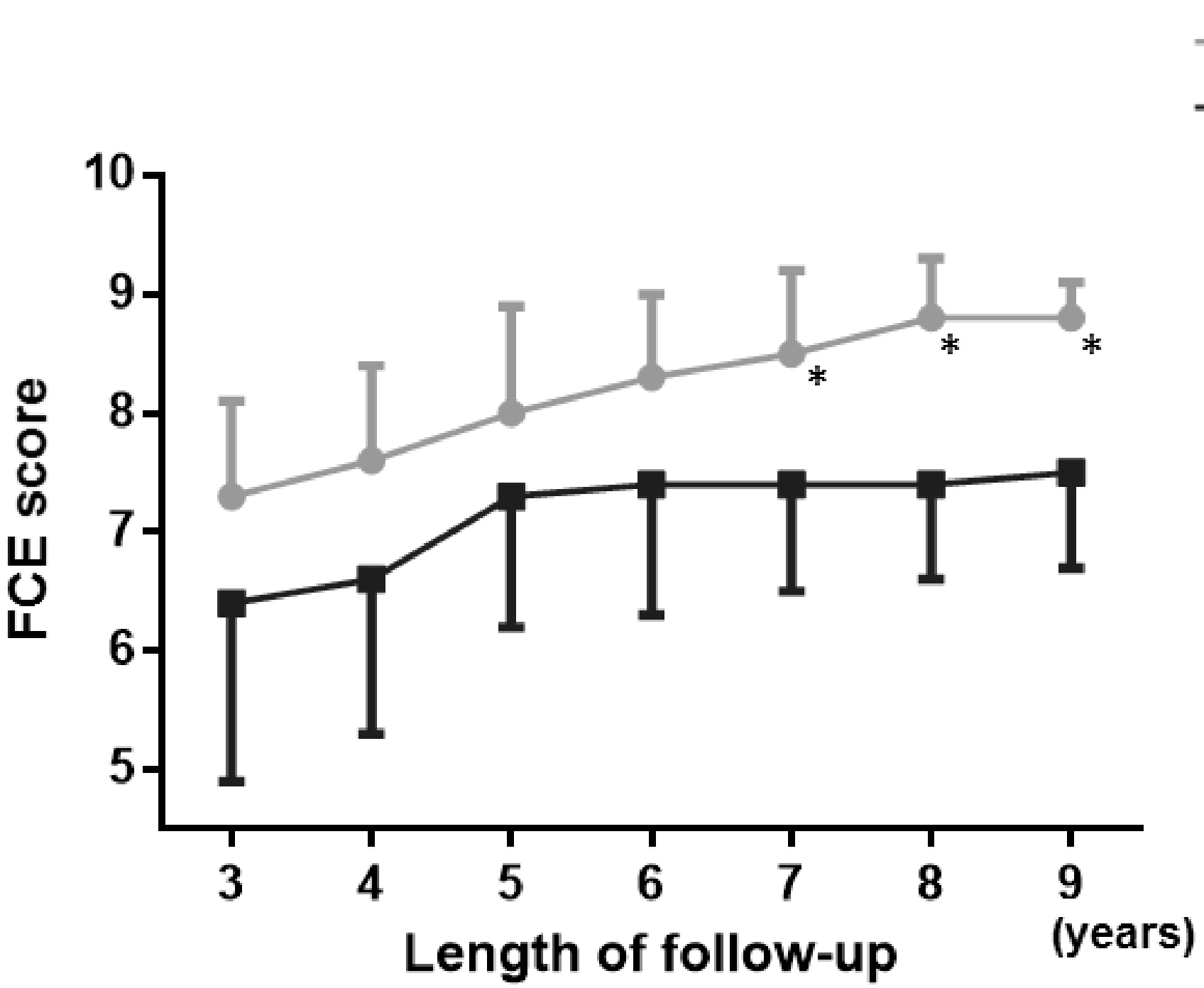
FIGURE LEGENDS

Figure 1: FCE score after LAARP and PSARP for RPF. Scores for LAARP are consistently higher and differences are significant at five, six and seven years after surgery (LAARP vs. PSRRP: 8.5±0.7 vs. 7.4±0.9 at five years after surgery, 8.8±0.5 vs. 7.4±0.9 at six years, and 8.8±0.3 vs. 7.4±0.7 at seven years, respectively).

Figure 2: FCE score after LAARP and PSARP for RBF. Scores for LAARP are consistently higher, but the differences are not significant.

Figure 3: FCESCV values are smaller in LAARP treated patients compared to those in PSARP treated patients for both RPF and RBF cases.


LAARP
PSARP
10
9
8
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FCE score
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3
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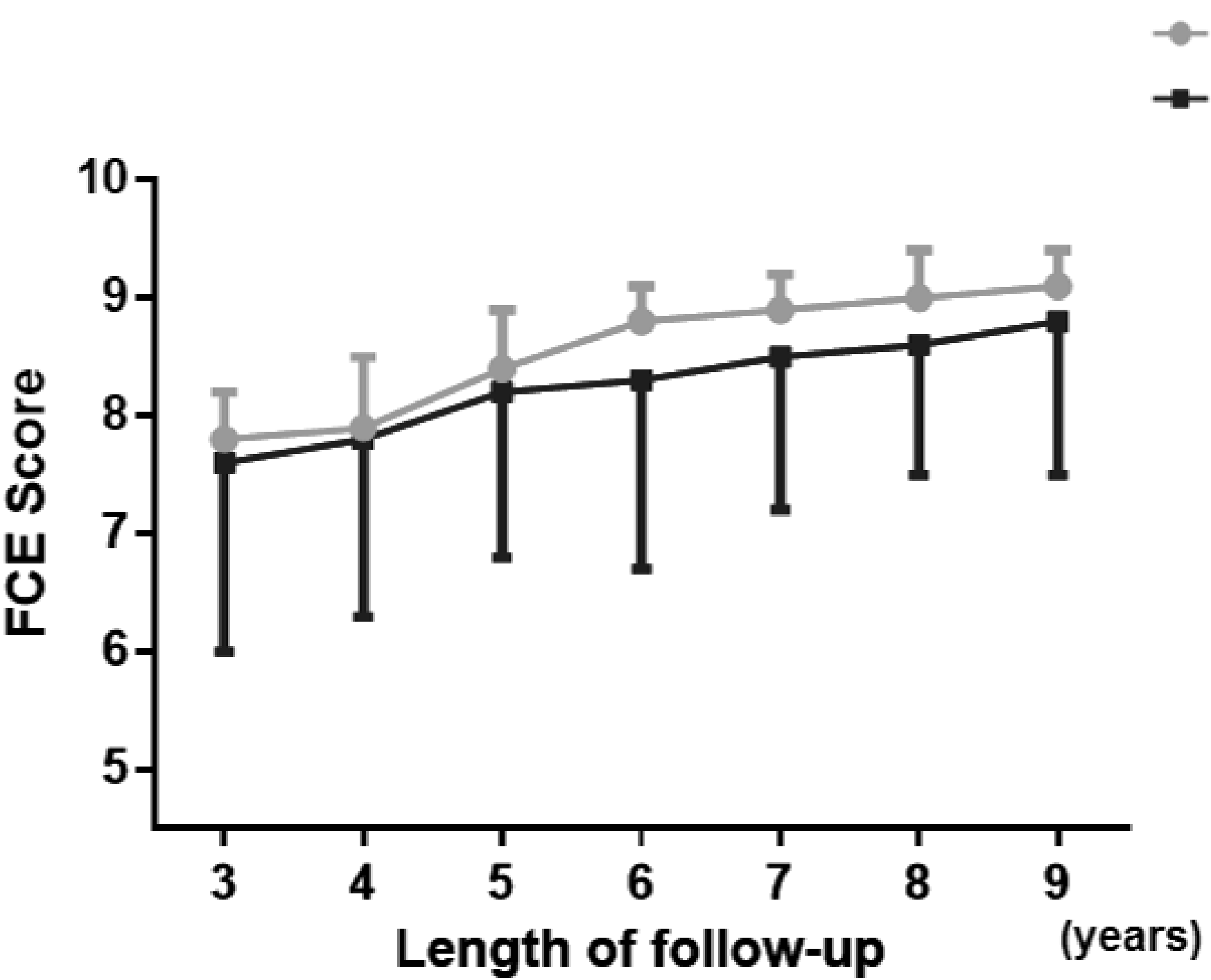
9
(years)
Length of follow-up


LAARP
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FCE Score
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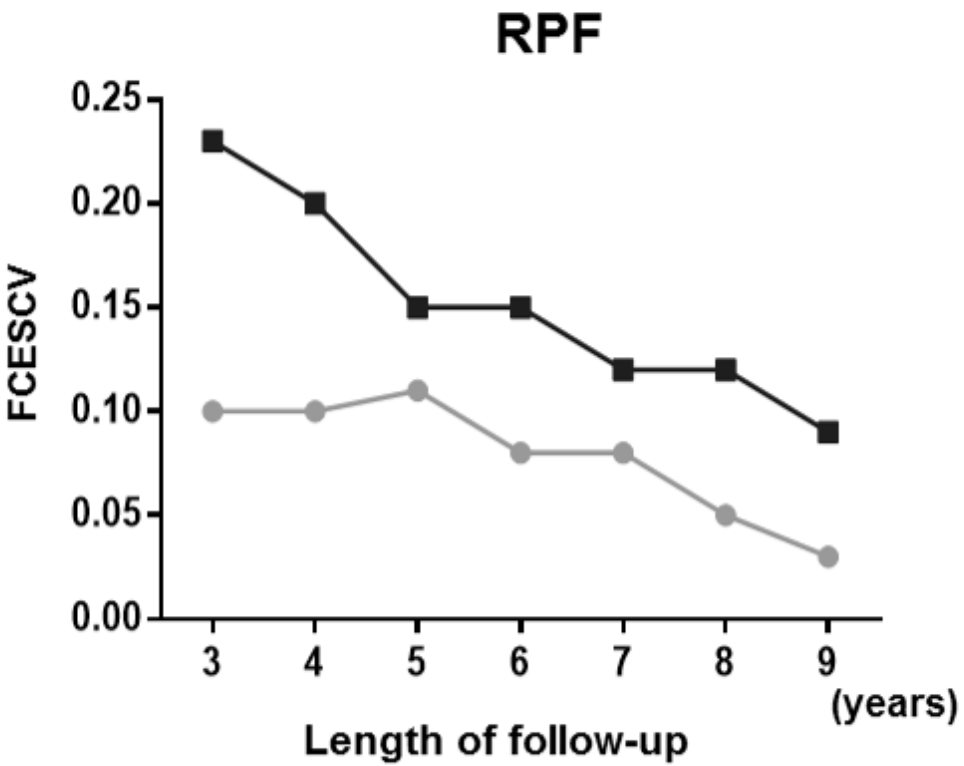
Length of follow-up
(years)


RPF
FCESCV
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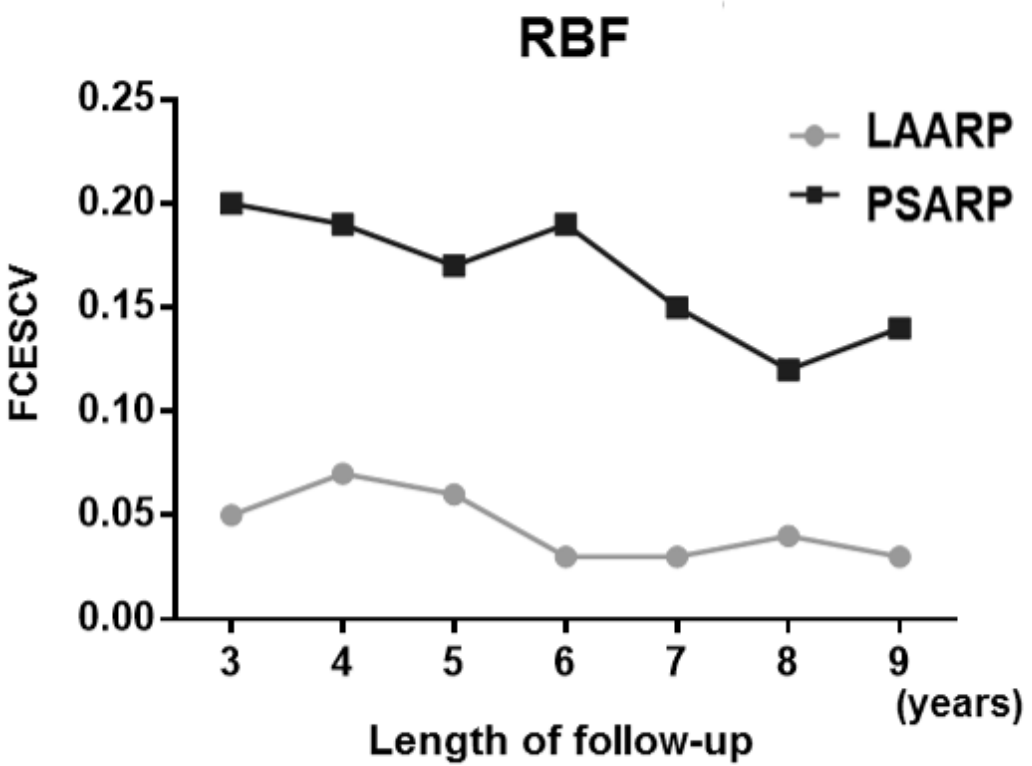
RBF
LAARP
PSARP
FCESCV
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Length of follow-up