

Long-term Follow-up After Surgery for Simple and Complex Cryptoglandular Fistulas: Fecal Incontinence and Impact on Quality of Life

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BACKGROUND: Surgical management of cryptoglandular fistulas is a challenge because the consequences of anal surgery potentially include fecal incontinence and impaired quality of life.

OBJECTIVE: To assess factors associated with fecal incontinence after surgery for simple and complex cryptoglandular fistulas and to determine the impact of incontinence on quality of life.

DESIGN: The design is retrospective and cross-sectional.

SETTINGS: This study was conducted at an academic tertiary center and at a private center specializing in proctologic surgery.

PATIENTS: All patients who underwent preoperative endoanal ultrasound for cryptoglandular fistula between 2002 and 2012.

MAIN OUTCOME MEASURES: A questionnaire was sent out in October 2013 to evaluate incontinence (Wexner score) and its impact on quality of life (FIQL). Variables

tested for association were patient demographics, fistula type, number of incised abscesses (0, 1, >1), number of fistulotomies (0, 1, >1) and number of sphincter-sparing procedures (0, 1, >1).

RESULTS: Of the 141 patients participating, 116 (82%; 76 men, 40 women) returned all the questionnaires. Median follow-up from the first perianal fistula surgery was 7.8 years (range, 2.1–18.1 years). Thirty-nine patients (34%) experienced incontinence. Surgical fistulotomy, multiple abscess drainages and a high transsphincteric or suprasphincteric fistula tract were associated with incontinence. As compared to simple fistula (Wexner score, 1.2 [SD, 2.1]), incontinence was worse after surgery for complex fistula (Wexner score, 4.7 [SD, 6.2], $p = 0.001$), as were quality of life elements, including lifestyle ($p = 0.030$), depression ($p = 0.077$) and embarrassment ($p < 0.001$).

LIMITATIONS: Mainly retrospective design without a standardized treatment protocol.

CONCLUSION: Surgical fistulotomy is the strongest risk factor for fecal incontinence. The severity of incontinence increases with the complexity of the fistula, negatively influencing quality of life. Special attention should be paid to these patients so as to mitigate symptoms later in life. A shift to sphincter-sparing procedures appears warranted.

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The surgical management of perianal fistulas remains a challenge because the consequences of anal surgery potentially include profound fecal incontinence (FI) and impaired quality of life (QOL).¹ The goal of surgical perianal fistula management is there-

fore to effectively eradicate anal sepsis while preserving anal function and continence. Fistulotomy (FT) in which the tract of the fistula is laid open is still considered the most effective procedure for structurally eradicating an anal fistula. However reported incidences of postoperative incontinence range from 4% to 62%.²⁻⁹ For higher and more complex fistulas, sphincter-sparing approaches such as core-fistulectomy or mucosal advancement flap are recommended as better approaches for preserving continence. Nonetheless, others report that laying open both low and high fistulas is associated with predictable rates of minor fecal incontinence^{8,10} In addition to the surgical treatment itself,^{4,11} risk factors that reportedly affect postoperative continence include age at surgery,¹² gender,^{4,13} fistula type and complexity,^{3,4,13,14} the presence of multiple fistula tracts,^{3,14} prior abscess drainage surgeries,⁷ and length of follow-up.¹⁵ Moreover, its embarrassing nature means FI is not always mentioned spontaneously, and data on long-term clinical outcomes after perianal fistula surgery are scarce. We postulated that if patients were specifically asked about FI, we would find that QOL is diminished to a greater degree in these patients than is generally believed. Therefore, the objectives of this study were to assess factors associated with FI after surgical procedures for simple and complex fistulas, and to determine the impact of resultant FI on daily QOL.

METHODS

A review of all patients who underwent preoperative 3D endoanal ultrasound (3D-EAUS) for a perianal fistula between 2002 and 2012 was performed at our tertiary center and at a local clinic specializing in proctologic surgery. At our tertiary center, 50% of patients with cryptoglandular fistulas underwent 3D-EAUS, and at the local clinic every patient underwent 3D-EAUS. Patients with noncryptoglandular fistulas were excluded. A fistula was considered to be of non-cryptoglandular origin when inflammatory bowel disease, a pilonidal sinus, hidradenitis suppurativa, tuberculosis, HIV, actinomycosis or anal carcinoma was also diagnosed. Patients who had undergone other sphincter surgery unrelated to an anal fistula, such as in anal sphincteroplasty or hemorrhoidectomy, were excluded. All patients included in the study underwent preoperative 3D-EAUS and concomitant fistula surgery resulting in closure of the fistula. Preoperative fistula classification and complexity was assessed during the 3D-EAUS in all patients, while information on the level and position of the fistulas in relation to anorectal anatomy was gathered through retrospective chart review. Fistula characteristics noted during the most recently performed 3D-EAUS were used. Fistula-closing operations performed for cryptoglandular disease were considered either anal sphincter-sparing or not. Core-fistulectomy¹⁶ and mucosal advancement flap were considered sphincter-sparing, as their principal goal

is to preserve the integrity of the anal sphincter. In cases of advancement flap placement, we always excised the fistulous tract by performed core-fistulectomy before placing the flap. In addition, a draining seton was placed 3–4 months before definitive advancement flap surgery was scheduled. Sphincter division through FT was defined as nonsphincter-sparing surgery. The number of abscesses surgically drained was also recorded.

3D-EAUS

Prior to surgery, 3D-EAUS was performed by a gastroenterologist (R. F.) or proctologic surgeon (C. D. G. Van der M.). Physical examination was performed with the patient in the left lateral decubitus position. A lubricated condom filled with ultrasonic gel was placed over the head of the probe, which was then gently introduced into the anal canal, just beyond the distal part of the rectum. In all patients, anal endosonography was performed using a 3D-EAUS system (Hawk type 2050, B-K Medical, Naerum, Denmark) with a rotating endoprobe housing 2 crystals covering 2–16 MHz (focal range, 2 to 4.5 cm; diameter, 1.7 cm) and producing a 360° view. While recording, the crystals were automatically pulled back by an internal puller, allowing longitudinal distances to be measured. Serial radial images and video recordings of the distal part of the rectum, puborectalis muscle, and internal and external anal sphincters were made. The fistula tract appeared as a hypoechoic tube-like lesion. When the presence of a complex fistula was suspected, 3% hydrogen peroxide was introduced into the fistula track using a flexible intravenous cannula. Infusion of hydrogen peroxide creates small air bubbles, illuminating the fistula track so that it appears bright hyperechoic on 3D-EAUS.

Classification

Fistulas were classified as subcutaneous, intersphincteric, low transsphincteric (involving the lower 1/3 of the external anal sphincter), high transsphincteric (involving the upper 2/3 of the external anal sphincter), suprasphincteric or extrasphincteric. A fistula was considered complex if there were multiple fistula tracts or a high transsphincteric, suprasphincteric or extrasphincteric fistula tract.

Questionnaires

In October 2013, questionnaires asking about current fistula-related perianal symptoms, current complaints of FI (Wexner score), and the impact of FI on QOL (FIQL) were sent out to patients. Current fistula-related complaints were defined as perianal pain, tenderness or fistula-related pus secretion in the preceding 4 weeks. If a patient did not return the first set questionnaires, they were sent a second set. To distinguish between fistula discharge symptoms

and FI, all patients were contacted by telephone by the author (A. P. Visscher). The follow-up period was the time in years from the first perianal fistula-related surgery up to October 2013. The Wexner score stratifies FI into flatus, liquid stool and solid stool, and objectifies the need for sanitary pads.¹⁷ The impact of FI on a person's QOL was measured using the FIQL, which has good psychometric properties and correlates well with FI severity.^{15,18} The FIQL is composed of 4 subscales: lifestyle, coping, depression and embarrassment. Scores range from 1 to 4, with a 1 indicating a lower functional status of QOL. This study was approved by the local Ethical Committee at the VU University of Amsterdam, the Netherlands.

Statistical Analysis

The variables analyzed were gender, age at first perianal sepsis-related surgery, perianal fistula type, number of perianal abscesses surgically incised and drained (0, 1, >1), number of FT's received (0, 1, >1) and the number of sphincter-sparing procedures received (0, 1, >1). The likelihood of each of these outcomes was estimated using univariate and multivariate logistic regression models, and the odds ratios (OR) and 95% confidence intervals (95% CI) were calculated. Variables from the univariate analysis with a *p* < 0.15 were included in the final multivariate logistic regression model. In the final multiple regression model, *p* < 0.05 was the criterion for statistical significance. In Table 4, the data on types of FI in patients with simple and complex perianal fistula were analyzed using the Fisher exact probability test, while the data on the severity of FI were analyzed using the t-test. In Figure 1, the impact of FI on QOL in patients with simple fistulas (SF) and complex fistulas (CF) was compared using the t-test. All analyses were performed using SPSS, version 20 (SPSS, Inc, Chicago, Illinois).

RESULTS

Written informed consent was obtained from all study participants. Of the 141 patients who met the inclusion criteria, 18 did not answer the questionnaires and 7 could not be reached due to incorrect contact information, which left 116 (82%) respondents for final analysis. The patient characteristics are listed in Table 1. Seventy-six (65%) patients were male. Among the 40 female participants, the mean parity was 1.9 (SD, 0.9; range, 0–4), and only 3 women had not borne children.

Incontinence

Median follow-up from the first perianal fistula-related surgery was 7.8 years (range, 2.1–18.1 years). Thirty-nine patients (34%) complained of FI. Table 2 shows the association between individual variables and FI. Univariate variables with a *p* < 0.15 were subsequently included in a

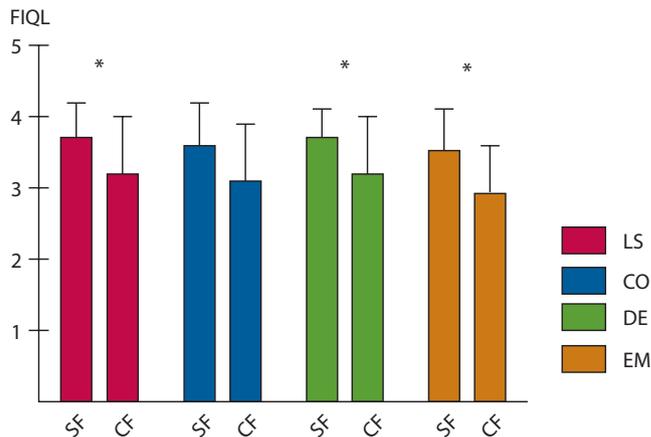


Figure 1. Mean FIQL scores in patients with FI (n = 40). FIQL, fecal incontinence quality of life score (0–4); LS, lifestyle; CO, Coping; DE, depression; EM, embarrassment; SF, simple fistula (n = 16); CF, complex fistula (n = 23). Scores from patients operated on for SF and CF were compared. Scores for LS (*p* = 0.030), DE, (*p* = 0.077) and EM (*p* < 0.001) were all lower in patients operated on for CF.

multivariate logistic regression model. Fistula characteristics, the number of abscesses incised, the number of FTs received and the number of sphincter-sparing procedures were associated with the presence of FI during follow-up. Patients with a subcutaneous fistula tract had a lower risk of FI than those with a suprasphincteric fistula. When >1 sphincter-sparing procedure had been performed, the risk for FI was greater than if no sphincter-sparing procedures were performed. However after adjusting for the other variables in the multivariate analysis, sphincter-sparing procedures were no longer associated with FI (Table 3). Patients who underwent >1 FT were at greater risk for FI than those who did not undergo FT. In addition, having a single FT in which >1 abscess was incised and drained or having a high transsphincteric or suprasphincteric fistula tract was also associated with FI.

Simple and complex fistulas

Of the 68 patients with SF, 57 (84%) required only 1 fistula closing operation. All 57 of those patients received

Table 1. Patient characteristics	
Characteristics	Values
Baseline	
Male: female, n (%)	76 (65): 40 (35)
Academic: private clinic, n (%)	72 (62): 44 (38)
Age at first surgery, y, median (range)	42 (17–73)
Surgery	
Fistulotomy, n (%)	87 (75)
Sphincter-sparing procedure, n (%)	53 (46)
Incision and drainage, n (%)	67 (58)
Follow-up	
Age at follow-up, y, median (range)	52 (27–83)
Fistula-related complaints, n (%)	4 (3)
Fecal incontinence, n (%)	39 (34)

Table 2. Variables associated with fecal incontinence

Patient characteristics	Complaints of FI, %	Univariate OR (95% CI)	p
Gender			
Female	34	[1]	–
Male	33	0.92 (0.41–2.04)	0.850
Age, y			
>40	33	[1]	–
<40	34	0.93 (0.43–1.93)	0.834
Fistula characteristics			
Submucous, n = 9	11	[1]	–
Intersphincteric, n = 41	32	3.70 (0.50–8.34)	0.232
Low transsphincteric, n = 21	27	3.03 (0.32–7.42)	0.345
High transsphincteric, n = 29	43	6.10 (0.74–17.22)	0.107
Suprasphincteric, n = 8	50	7.01 (0.62–35.10)	0.120
Extrasphincteric, n = 8	40	5.32 (0.31–21.34)	0.232
Fistula surgery, no. I & D			
0	20	[1]	–
1	40	2.63 (0.94–4.90)	0.053
>1	47	3.41 (1.42–8.40)	0.014
No. FT			
0	23	[1]	–
1	30	2.91 (0.73–4.42)	0.291
>1	80	14.84 (3.22–34.10)	<0.001
No. SSP			
0	24	[1]	–
1	32	1.82 (0.83–4.14)	0.151
>1	60	4.30 (1.23–13.10)	0.019

Variables tested for association with the presence of FI. Univariate logistic regression analysis was performed, and the percentage of patients with complaints of FI is shown per variable.

Age = age of first perianal fistula operation; I & D = incision & drainage; FT = fistulotomies; SSP = sphincter-sparing procedures.

a FT; of those, 9 (13%) had undergone >1 (range 1–3) FT. Seven (10%) patients received a sphincter-sparing operation; of those, 1 had undergone >1 such procedure. Among the patients receiving FT, 36 (53%) had undergone abscess incision and drainage, and 14 (21%) had undergone >1 drainage.

Of the 48 patients with CF, 19 (40%) required only 1 fistula closing operation. Forty-five (94%) had received a sphincter-sparing procedure, of which 19 (40%) had undergone >1. In addition, 19 (40%) patients with CF underwent a FT; of those, 6 (13%) had undergone >1 (range 1–2). Among patients receiving a sphincter-sparing procedure, 31 (65%) had undergone abscess incision and drainage, and 18 (38%) had undergone >1.

Whether or not patients with SF or CF experienced FI at least once per month is shown in Table 4. The table also reports the severity of FI based on the Wexner incontinence score, with higher scores indicating more severe complaints and more frequent episodes of FI. FI involving solid stool, gas and the need for sanitary pads was more severe among patients who had surgery for CF than among those operated on for SF (Wexner score: SF, 1.2 (SD 2.1) vs. CF, 4.7 (SD 6.2), $p = 0.001$).

Table 3. Variables associated with fecal incontinence

Fistula characteristics	Multivariate OR (95% CI)	p
Submucous	[1]	–
Intersphincteric	1.72 (0.21–3.40)	0.691
Low transsphincteric	1.41 (0.52–7.14)	0.835
High transsphincteric	2.81 (1.50–16.93)	0.032
Suprasphincteric	4.13 (2.12–42.80)	0.011
Extrasphincteric	7.12 (0.71–34.80)	0.066
Fistula surgery, no. I & D		
0	[1]	–
1	1.63 (0.70–4.63)	0.134
>1	3.12 (1.53–7.80)	0.021
No. FT		
0	[1]	–
1	2.92 (1.10–18.64)	0.001
>1	9.44 (1.81–31.90)	<0.001
No. SSP		
0	[1]	–
1	1.43 (0.40–4.81)	0.253
>1	3.24 (0.72–8.32)	0.114

Multivariate logistic regression analysis was performed. $p < 0.05$ was considered statistically significant.

FT = fistulotomies; I & D = incision & drainage; SSP = sphincter-sparing procedures.

QOL

All patients with FI stated that it negatively affected their QOL in some degree. The 4 patients with both FI and fistula-related perianal complaints all stated that FI impaired their QOL. Those patients were contacted by telephone and asked to visit our academic center for physical examination, and all 4 reported anal tenderness or pain without active pus secretion. Lifestyle and everyday behavior were negatively affected in 21 (18%) and 25 patients (22%), respectively. Feelings of depression were reported by 27 (23%) patients, and embarrassment by 29 (25%). All 4 subcategories of the FIQL were scored lower by patients with FI ($p < 0.001$). Mean FIQL subscales for lifestyle ($p = 0.030$), depression ($p = 0.077$) and embarrassment ($p < 0.001$) were all scored lower by patients operated on for CF than by those treated for SF (Fig. 1).

DISCUSSION

Possible complications of perianal fistula-related surgery include recurrence and FI. Therefore, the main goal of surgical treatment is structural fistula closure without postoperative FI. Delineating the precise fistula pattern is mandatory for effective eradication of all septic material. We have found preoperative 3D-EAUS to be a helpful tool that provides the surgeon with important information about the specific anatomy of a patient's perianal fistula. Furthermore, preoperative identification of fistula- and patient-specific characteristics associated with postoperative FI is important, as they can influence a surgeon's judgment regarding what treatment will best preserve sphincter functionality.

Table 4. Type and severity of fecal incontinence at final follow-up

Type of incontinence	Total (n = 116)	SF (n = 68)	CF (n = 48)	p
Solid stool				
Yes (%) / No (%)	13 (11) / 103 (89)	2 (3) / 66 (97)	11 (23) / 37 (77)	0.002
Liquid stool				
Yes (%) / No (%)	7 (6) / 109 (94)	2 (3) / 66 (97)	5 (10) / 43 (90)	0.124
Gas				
Yes (%) / No (%)	39 (34) / 77 (66)	16 (24) / 52 (76)	23 (48) / 25 (52)	0.009
Needing sanitary pads				
Yes (%) / No (%)	17 (15) / 99 (85)	3 (4) / 65 (96)	14 (29) / 34 (71)	0.001
Wexner score				
Mean (SD)	2.5 (4.6)	1.2 (2.1)	4.7 (6.2)	0.001

Shown are the numbers of patients reporting the indicated type of incontinence at least once per month. The Wexner incontinence score was used to assess the severity of fecal incontinence, with higher scores indicating greater severity.

CF = complex fistulas; SF = simple fistulas.

Most studies of perianal fistulas focus almost exclusively on clinical outcomes; few evaluate the impact of clinical outcome on daily perception of health and QOL. This long-term follow-up study examined the factors associated with FI and assessed the impact of FI on QOL after surgery for simple and complex anal fistulas. We found that a high trans- or suprasphincteric fistula tract, more than a single surgical incision or drainage of a perianal abscess, or having received at least one FT was associated with FI. Fistula type as a risk factor for FI may be explained physiologically as reflecting the loss of muscle mass after surgical intervention. Furthermore, high trans- and supra-sphincteric fistulas tend to persist and recur more often than simpler, more distally located fistulas. Consequently, they are more likely to be associated with repetitive surgical interventions, which further increases the risk of FI. Consistent with this interpretation is the finding that more than one surgical abscess incision was also associated with FI. Repeated drainage damages small nerves and creates more scar tissue around the anorectum.³ In addition, repeated abscess formation and prolonged inflammation may also damage the anal sphincters, further impairing their function. However our study did not differentiate between repetitive drainage of a single abscessed area and abscess drainage of different areas with years in between.

The strongest risk factor for FI was whether or not more than one FT had been performed. In our study all patients with SF and 40% of those with CF had undergone FT, with more frequent and more severe FI in patients with CF. Management of anal fistulas using FT is a delicate balance between cure and continence, especially with higher more complex fistulas.¹⁹⁻²² The risk of postoperative FI is believed to be minimal if less than 33% of the lower external anal sphincter is penetrated. In these cases, FT is recommended.^{3,4}

A follow-up study by Jordan et al evaluated FI in 279 patients (43% CF) 5 years after anal fistula surgery.²² FI was present in 8% of patients with SF and in 18% of those with CF. FT had been performed in 132 patients, 95% for simple intersphincteric or low transsphincteric

fistulas. Patients with CF were predominantly treated using core-fistulectomy and closure of the internal opening, core-fistulectomy and sphincter reconstruction, or core-fistulectomy and mucosal advancement flap. FT had a higher associated risk of FI than any of those. Another study by Bokhari et al²¹ compared the outcomes of FT and sphincter-saving procedures in 128 patients with perianal fistulas. They found frequent (49%) use of FT in CF, with 37% of patients reporting FI. Patients who underwent FT for CF also reported higher rates of major FI (13%) than patients who underwent a sphincter-saving procedure (0%). Nevertheless, some report that FT can be performed for higher more complex fistulas.^{8,10,23} A 3D-EAUS study by Garces-Albir et al reported that division of the lower 66% of the external anal sphincter in FT was associated with excellent continence and cure rates in patients who lacked risk factors before surgery.²³ Patients with major FI, inflammatory bowel disease or prior anal fistula surgery, or female patients with anterior transsphincteric tracts or obstetric anal sphincter injuries were excluded. Among the 36 patients they studied, the recurrence rate after 1 year was 0%, and FI scores before and after surgery did not differ. However, follow-up was short and FI may become a more serious problem later in life, as the capacity of the compensatory continence-preserving mechanisms declines.²⁴

We found a significant reduction in QOL in patients with FI, with more severe and invalidating complications in patients operated on for CF. In a large study of 624 patients who had received surgical treatment for anal fistulas, it was concluded that recurrence, the presence of FI and the effects of FI on daily life were all associated with patient satisfaction.²⁵ Moreover, the greatest amount of dissatisfaction was attributable to the presence of FI and its impact on daily life. Another study using the FI severity index to assess FI after FT reported that the amount of external anal sphincter division was the only variable associated with FI severity.¹⁴ Furthermore, FI severity correlated strongly with reported QOL. So is it possible to avoid postoperative FI through careful patient selection? Elderly pa-

tients and multiparous females are at higher risk, whereas males under 60 have stronger sphincters and are thus at lower risk.

Prospective manometric studies report that most fistula operations negatively affect anal function and continence.^{6,13,16,26–28} The degree of deterioration appears acceptable after FT for low intersphincteric⁶ and transsphincteric tracts,¹³ except when preoperative FI or low anal pressures are present. Because FT for higher more complex fistulas is much more likely to impair anal function,^{13,16} a shift to more sphincter-sparing procedures is warranted. Functional results of the recently introduced ligation of the intersphincteric fistula tract procedure (LIFT) are promising,²⁹ though we are awaiting long-term results. For patients with complex recurrent anal fistulas and concomitant FI, the results of combined FT and sphincter reconstruction show better function and clinical outcomes.^{9,30}

This study finds that FI is a debilitating problem that persists long after surgery to treat perianal fistula has been performed. However, because complaints of FI tend to worsen over time, there is room for potential bias, as we included patients followed up for differing periods of time. Nevertheless, we believe that patients eligible for perianal fistula surgery should not only be informed of their risk of postoperative FI, but also of the potential adverse effects FI may have on their lifestyle and emotional stability. Additionally, patients undergoing surgery for more complex fistulas may be more severely affected by FI and its impact on daily life. FI rates were lower in patients who underwent sphincter-sparing procedures, and a shift to performing more such procedures is warranted. That said, more research is needed on the efficacy and sustainability of the different sphincter-sparing procedures. This study should be considered in the context of its limitations, which are its mainly retrospective design and the absence of a standardized treatment protocol.

CONCLUSION

In this follow-up study, surgical FT was the strongest factor associated with the presence of FI, and the severity of FI increased with the complexity of the perianal fistula tract, negatively affecting QOL. Special attention should be paid to these patients so as to mitigate symptoms later in life. A shift to performance of more sphincter-sparing procedures for eradicating perianal fistulas is warranted.

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